



Feetus papyraceous in a monoamniotic monochorionic pregnancy: a case report.

Ruchi Sinha, Sonali Gupta, Shikha Sachan, Anuradha Khanna

Department of Obstetrics and Gynecology, Institute of Medical Sciences, BHU, Varanasi 221005
India

CASE REPORT

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Corresponding Author:

Dr. Ruchi
7, Saryoonagar, Kamachha, Varanasi-221010
U.P. (INDIA)
dr.ruchi_bhu@yahoo.com

Abstract

Foetus papyraceous or compress 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. Incidence of foetus papyraceous in twin pregnancy is 1 in 184 to 1 in 200 pregnancies. A case of foetus papyraceous which was discovered following expulsion of a Twin at 17 weeks and 1 day of gestation with low lying placenta in a monochorionic monoamniotic twin pregnancy is reported here.

Key Words: foetus papyraceous, intrauterine death, monochorionic monoamniotic twin

Background

Foetus papyraceous or compress is the compressed, mummified, parchment-like remains of a dead twin which is retained in-utero after intrauterine death in the second trimester.^{1, 2} It is usually discovered among the placenta and membranes of its well developed twin. The cause is thought to be death of one twin, amniotic fluid loss, or reabsorption and compression of the dead foetus. The incidence of foetus papyraceous has been reported at 1 in 17,000 to 20,000 pregnancies and 1 in 184 to 1 in 200 twin pregnancies.^{2, 3}

A 20 years old primi gravida referred to S.S. hospital, BHU, India with ultrasound (USG) diagnosed intrauterine dead (IUD) twin pregnancy at 17 weeks and 1 day of gestation. She was having per vaginal (P/V) bleeding for 18 days. There was history of occasional on and off P/V bleeding in the present pregnancy since 3 months of gestation. There was no history of expulsion of product of conception. Her menstrual cycles were regular at an interval of 20 days and lasting for 3-4 days. Her previous ultrasound report done one day back showed twin pregnancy at 14 weeks and 6 days of gestation. Cardiac activity was absent in both the foetuses indicating intrauterine death. The first was cephalic and second was breech and the placenta was low lying.

On examination the patient was severely pale but her vitals were stable.

Obstetrical examination revealed that the fundal height was more than the period of gestation multiple foetal poles were palpable and foetal heart sound could not be localized by Cardiotocography (CTG) probe. Uterine contractions were of moderate intensity lasting for 20-25 sec at 15-20 minutes interval. Speculum examination showed presence of around 500-750 ml blood clots in the vagina. The cervical os was open and there was active bleeding. The patient's blood group was O positive and haemoglobin level 5.5 gm%. Her coagulation profile was normal. To correct the haemoglobin level immediately 2 units blood was transfused. The first foetus, male, 40 gms in weight which expelled spontaneously, was compressed, mummified, parchment like, suggestive of foetus papyraceous. The second twin, male weighing 225 gms, expelled immediately after and was plethora (Fig.1).

On examination of the placenta it was found to be monoamniotic and monochorionic (Fig.2, 3). The histopathology report of placenta confirmed this and there was no twin to twin transfusion syndrome. The umbilical cords were normal.

Oxytocics were given and P/V bleeding was normal. The uterus was well contracted. Two more units of blood were transfused in the postpartum period. The patient received antibiotics and was discharged on the third postpartum day. The patient came for follow up after 2 weeks.



Discussion

Twin pregnancy or multiple gestation is a common finding in today's era especially due to the ART and IVF.⁴ Stoeckel (1945)⁵ was the first to suggest that the conception rate of multiple gestations was greater than the birth rate. Hence, the death of foetuses in a 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.⁵

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.^{1,2} Frequently the only sign left of vanishing twin is the development of subtle abnormality of placenta such as well-defined cysts or sacs,⁶ areas of degenerated chorionic villi,⁷ fibrin deposition or fibrinoid degeneration,⁸ placental nodules or plaques and embryonic remnants.⁸ Causative factors for foetus papyraceous have been debated in literature. The role of velamentous and marginal insertion of placenta has been postulated.⁹ The condition occurs more often in foetuses with genetic or chromosomal abnormalities.⁵ Placental or foetal analysis frequently reveals diploidy, triploidy, and alternate sex chromosome on placental biopsy, foetal skin biopsies, and chorionic villous sampling.^{5, 10, 11} Rare case of a foetus papyraceous due to maternal trauma has also been reported in the literature.⁴

Earlier, foetus papyraceous was thought to be more common in monozygotic twins.¹² But recently Benson et al showed similar frequencies in dichorionic (12%) and monochorionic twin (11%)¹³ pregnancies however, there were only nine monochorionic twins in the study. Ours was a case of monochorionic twin pregnancy. It is more difficult to predict viability of twins early in pregnancy, more so when monochorionic.¹³

There is no prognostic effect of maternal age and spontaneous versus induced conception.¹⁴

The primary concern of foetus papyraceous is its effect on mother and surviving co-twin. In many cases no complications to the mother or to the surviving twin have been reported.¹⁵ 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.⁵ 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.¹⁵ Vanishing twin in a pregnancy significantly increases both preterm (<37 gestational weeks) and very preterm (<32 gestational weeks) births.¹⁵ 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.⁹ In the case presented, the twin was plethoric.

Anand et al¹⁶ reported that surviving co-twins had poorer scores on the Griffiths Mental and Development Scales when compared to singleton pregnancies. 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 intravaginal probe.² The subsequent demise of one foetus could then be diagnosed before delivery. In this case, however, mid trimester ultrasound failed to diagnose papyraceous twin. 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).¹⁷ Alpha-fetoprotein levels are elevated compared with values at similar junctures in both singleton pregnancy and normal twin pregnancy.¹⁸ The rate of rise of beta-hCG is slower than that in a normal twin pregnancy.¹⁹

As technology advances, the routine surveillance of early pregnancy will be helpful in early detection of foetus papyraceous before the development of complications.

Conclusion

Foetus papyraceous although rare is a well known entity. It is very important to make a diagnosis in time to prevent severe complications. An ultrasound proves to be of great help in making the diagnosis.

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PEER REVIEW

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CONFLICTS OF INTEREST

The authors declare that they have no conflict of interest.

Fig. 1. Foetus papyraceous and other plethoric co-twin



Fig.2 Foetal surface of placenta



Fig.3 Maternal surface of placenta





Fig. 4 Foetus papyraceous, the other twin and the placenta

