

A Rare Sequela of Twin Demise: Fetus Papyraceous

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ABSTRACT:

Fetus papyraceous is a rare condition where there is intrauterine fetal compression of one of the fetuses of a twin pregnancy, which may cause complete resorption or result in a parchment paper-like appearance of that fetus. We report one such case of twin pregnancy in a 23-year-old primigravida with 36 weeks of amenorrhoea. Ultrasonography done during her antenatal visits revealed a twin pregnancy with one dead fetus. She delivered one healthy baby, the placenta, and another pale white membranous tissue with firm areas. Histopathological examination of multiple tissue sections of the specimen revealed placental tissue characterised by firm consistency, greyish discolouration, and areas of calcification. Fragments of an umbilical cord were also observed. Additionally, a markedly compressed fetus was identified, exhibiting significant limb deformities, confirming the diagnosis of fetus papyraceous. The basic concern of a fetus papyraceous is its detrimental effects on the mother and the surviving fetus. To avoid possible complications, intrauterine diagnosis of a fetus papyraceous by serial ultrasound examinations and routine placental examination post-delivery should be keenly followed. The pregnancy was diagnosed as a dichorionic diamniotic twin gestation with intrauterine demise of one fetus during the second trimester.

Keywords: *Fetus papyraceous, Twin pregnancy, Vanishing twin syndrome, Intrauterine fetal demise, Histopathology, Placenta*

INTRODUCTION:

A Fetus papyraceous or fetus compressus is a rare complication of multiple pregnancy resulting from the intrauterine demise of one fetus, with retention of the dead fetus for a prolonged period [1,3]. The dead fetus becomes compressed between the uterine wall, membranes, and placenta of the surviving co-twin, leading to dehydration, mummification, and flattening into a thin, parchment-like fetal remnant known as fetus papyraceous. Initially, a multiple pregnancy may be diagnosed in early gestation, but serial ultrasonographic examinations may show the disappearance of one or two twins. This phenomenon is sometimes referred to as vanishing twin syndrome [4]. Placental blood vessels form anastomoses, resulting in blood being redirected from one twin (the donor) to the other [6,9]. The donor twin develops anaemia, restricted growth, and oligohydramnios. If this process occurs early in gestation, typically around the third or fourth month, the affected fetus may be compressed and flattened by the surviving co-twin due to incomplete ossification of the

trunk and the gradual resorption of fluids from the tissues. Complete resorption of the fetus may present as first-trimester bleeding. However, if this event arises during the second or third trimester, it can give rise to significant complications for the mother and surviving fetus, such as missed abortion, pre-eclampsia, maternal infection, DIC, thrombotic thrombocytopenic purpura, intrauterine fetal demise, premature babies, low birth weight babies, and hypoxic ischaemic encephalopathy [5,7,8]. In late pregnancy, a fetus papyraceous situated low in the uterus may obstruct the cervical canal, potentially necessitating caesarean section for the delivery of the surviving twin [2,12].

The occurrence of fetus papyraceous is estimated to be approximately 1 in 17,000 to 20,000 pregnancies [1,2]. Among twin pregnancies, this intrauterine event is observed in roughly 1 out of every 184 cases (0.54%), while in triplet pregnancies, the incidence is about 1 in 8,000 or approximately 1 in 32,800 for triplets specifically. The presence of two papyraceous fetuses in a triplet gestation is uncommon. Although fetus

papyraceus may develop in both monozygotic and dizygotic twins, it is more frequently seen in monozygotic twins[5,9]. Its incidence has increased over the past few years due to the use of assisted reproductive techniques [4,8].

CASE REPORT:

A 23-year-old primigravida with 36 weeks of amenorrhoea was referred to the obstetrics and gynaecology outpatient department for planning of delivery. She had taken regular antenatal visits and had undergone periodic ultrasounds at a rural health centre. Her first trimester ultrasound at the 9th week revealed a dichorionic and a diamniotic twin gestation. Another ultrasonography taken during her early second trimester, at 16 weeks of gestation, showed one live fetus of 16 weeks 3 days, and another fetus showed

demise with a gestational age of 14 weeks 5 days. Subsequent ultrasounds showed adequate development of one fetus and a retained dead fetus. At the gestational age of 32 weeks 2 days, an echogenic focus was seen to be adherent to the decidua of the live fetus, which was reported as tissue of fetal remnants of the deceased twin. During the time of admission to our hospital, at 37 weeks 3 days, her general physical examination was normal, with a APTT was 39.5 seconds (reference range 25–35 seconds) and D-dimer was 1.2 µg/mL. She was conservatively managed with a close watch on her coagulation profile. She delivered a healthy baby weighing 2800 g by normal vaginal delivery. The term placenta was delivered along with a small pale white membranous tissue with firm areas, which was sent for histopathological examination.

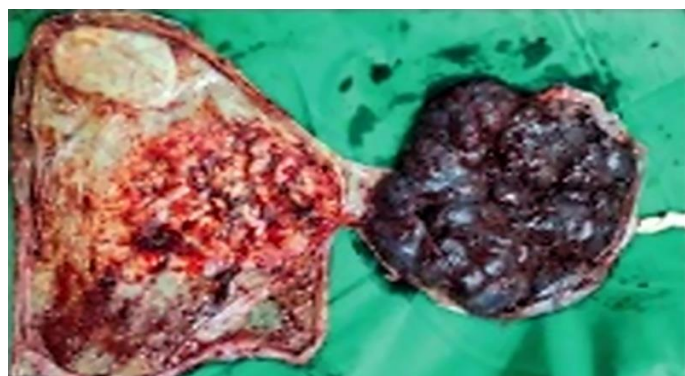


Figure 1: Fetus papyraceus and placenta

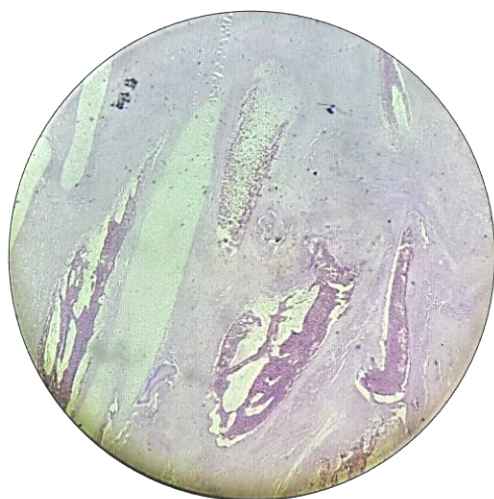


Figure 2: Villous fibrosis and calcifications, H&E stain, and original magnification 10x

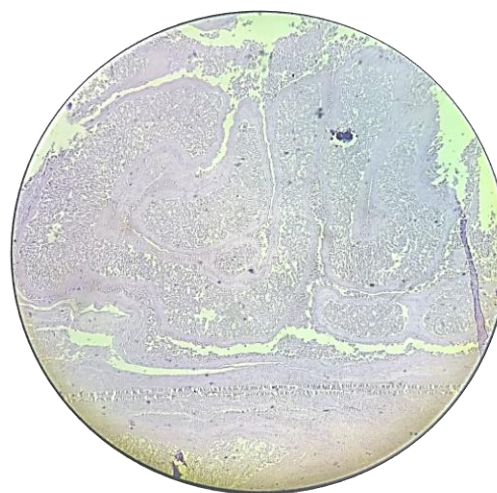


Figure 3: Fibrous change in the placental stroma with focal necrosis, fibrin extravasation and haemorrhage, H&E stain, and original magnification 10x

EXAMINATION:

On gross examination, the pale whitish tissue received along with the placenta measured 12 cm x 10 cm x 1.5 cm, having predominantly thin parchment-like areas and few foci appearing firm. Multiple sections were taken from firm and thin areas. On microscopy, sections through the tissue showed areas of developing bone, cartilage, ciliated epithelium, and muscular tissue,

confirming the diagnosis of fetus papyraceus. Histopathological examination also revealed placental tissue characterised by firm consistency, greyish discolouration, and areas of calcification. Fragments of an umbilical cord were also observed. Additionally, a markedly compressed fetus was identified, exhibiting significant limb deformities.

DISCUSSION:

Fetus papyraceous represents one outcome of vanishing twin syndrome when fetal demise occurs after sufficient fetal development and the demised fetus remains compressed within the uterine cavity[4]. Due to the prevalence of vascular connections (85–98%) in cases of uniovular twins, intrauterine death is three times more common[9]. Maternal complications include preterm labor, consumptive coagulopathy, labor dystocia, and sepsis as a result of the retention of a dead fetus[5,7]. Surviving fetal complications include congenital malformations, intrauterine growth restriction, prematurity, and death[8,10]. Gastroschisis, aplasia cutis, intestinal atresia, central nervous system damage, absent ears, and anomalies of the heart are the congenital anomalies reported so far in the live fetus[10,11]. These anomalies are mostly caused by thrombus and other clotting factors released from the dead fetus, which embolizes the live twin to produce vascular occlusion[9,11].

If the fetus is absorbed completely in the first trimester, there are usually no further complications to the pregnancy. Our patient was diagnosed with fetal papyraceous early in the second trimester due to good diagnostic facilities. The complication can be severe in a monochorionic placenta rather than a dichorionic placenta[8,9,10]. No association of parity or maternal age was found. In most cases, both the mother and the surviving twin remain healthy with no complications, as in our case[2,12].

To assess the consumptive coagulopathy and maternal infection, biochemical, haematological, and ultrasonography monitoring of the mother in the antenatal period as well as after delivery is very important[5,8]. Detailed newborn examination is important to rule out congenital malformations. Histopathological examination of the placenta in the postpartum period can be helpful in knowing the causes of fetus papyraceous[1,3,12].

Ethical approval:

Ethical approval was waived as this is a single case report and written informed consent was obtained from the patient.

Learning Points:

- Fetus papyraceous is a rare complication of multifetal gestation.
- Early ultrasonographic diagnosis is essential.
- Serial maternal coagulation monitoring should be performed.
- Histopathological examination confirms the diagnosis.

Limitations:

The absence of serial coagulation profile measurements and limited imaging documentation of the demised fetus represent limitations of the present case report.

CONCLUSION:

In twin or multiple pregnancies, after delivery, the placental tissue should be thoroughly examined, and if found suspicious, should be sent for histopathological examination for confirmation of fetus papyraceous. The main concern of a fetus papyraceous is its impact on the surviving fetus and on the mother; serial ultrasound examinations and regular monitoring of the mother are recommended to avoid any possible complications. To avoid possible complications, intrauterine diagnosis of a fetus papyraceous by serial ultrasound examinations and routine placental examination post-delivery to search for a fetus papyraceous should be keenly followed.

DECLARATION OF PATIENT CONSENT:

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

Conflict of Interest:

The authors declare no conflict of interest.

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Author Contributions:

MM collected clinical data, performed literature review and drafted the manuscript. MTG supervised the study, interpreted histopathological findings and critically revised the manuscript. Both authors approved the final manuscript.

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