

Fetus Papyraceus: a rare case report

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***Keywords:** Fetus Papyraceus, Multiple Pregnancies, Intrauterine Death.*

Abstract

Fetus papyraceus (FP) also called fetus compressus is a mummified and compressed fetus as a result of fetal death during multiple pregnancy. Fetus papyraceus is a rare condition and is associated with obstetric complications. Sonography is a visual tool to confirm the diagnosis, but it is not always possible due to anatomical position and technical difficulties. FP is usually discovered among the placenta and membranes of its well developed twin. Searching for a fetus papyraceus should be a routine part of placental examination.

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Date of Submission: July 3, 2014

Date of Acceptance: Oct 3, 2014

Date of Publishing: Jan 28, 2015



Introduction

Fetus papyraceus or Fetus compressus is the compressed, mummified, parchment like remains of a dead twin which is retained in utero after intrauterine death in second trimester.^[1] It is a macerated, tiny, fully formed fetus which is usually dry and papery because the amniotic fluid, fluid content of the dead fetal tissues and of the placental tissue gets absorbed and the dead fetus gets flattened and compressed between the membranes of the living co-twin and the uterine wall.^[2] It is relatively a very rare complication in twin pregnancy.^[3] The incidence of fetus papyraceus has been reported as 1 in 17,000 to 20,000 pregnancies. Incidence of fetus papyraceus in twin pregnancy is 1 in 184 to 1 in 200 pregnancies.^[1] FP can occur in both uniovular and binovular twins but is more common in uniovular twins.^[4] The cause is thought to be death of one twin, amniotic fluid loss, or reabsorption and compression of the dead fetus. Death of one twin in first trimester with vanishing twin syndrome is relatively common (upto 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).^[1] Here we present a case of twin pregnancy of a 23 year old female with singleton normal fetus with fetus papyraceus formation of other twin.

Case Report

A 23 year old female patient (G₂P₁L₁) with 6 days post date with twin gestation came in active labour to our hospital. On admission, she had irregular uterine contractions, per vaginal examination showed fully dilated cervix with intact membranes. All laboratory tests were within normal limits. Two USG reports done during 18th and 31st weeks revealed dichorionic diamniotic gestation with early fetus papyraceus formation of the other twin. She delivered a healthy male child weighing 2900 gms by normal vaginal delivery. The term placenta was delivered along with a second small pale white placenta. The two placentae were sent for histopathological examination.

Gross examination: The larger term placenta measured 15x12x4 cms and weighed 650 gms. It showed marginal insertion of the umbilical cord. The external and cut surfaces appeared unremarkable (Fig 1). The other placenta was small, discoid, pale white to tan, the cut surface showed whitish appearance with few

dark brown areas (fig 2). Grossly there was no fetus papyraceus evident however on radiograph of this smaller placenta showed two clavicles, vertebral column and rib cage of fetus papyraceus (fig 3). In our case, the fetus papyraceus was compressed and embedded in the placental tissue.



Figure 1. Larger placenta with marginal insertion of cord and smaller placenta appears discoid grey white mass.



Figure 2. Cut surface of smaller placenta, revealing grey white area with few hemorrhagic areas.

Microscopy: Sections through the smaller placenta showed developing vertebral column of fetus papyraceus along with developing gastrointestinal tract, kidney and skeletal muscle adjacent to it. Extensive perivillous and intervillous fibrin deposition with entrapped chorionic villi showing fibrosis were noted. Occasional sheets of deciduas showed focal dense

infiltration by polymorphs and mononuclear cells (fig 4).

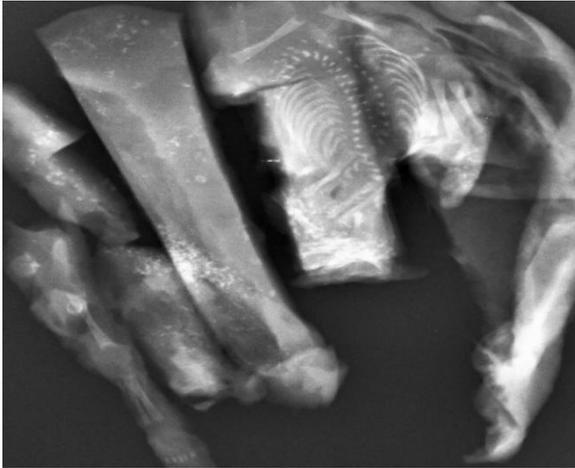


Figure 3. Radiographic picture of small placenta, revealing skeletal system of fetus papyraceus, comprising of vertebral column, rib cage, clavicles and thigh bones.

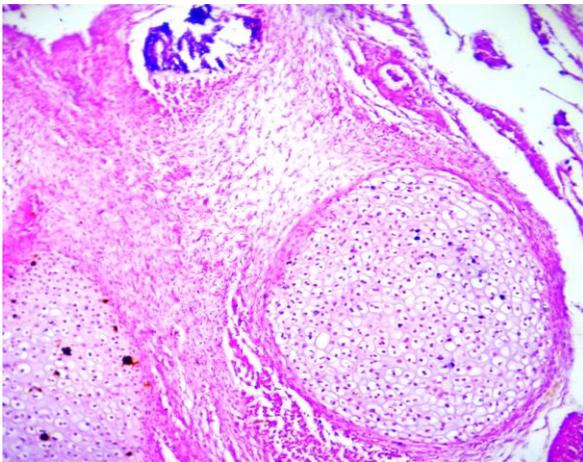


Figure 4. Microscopy of small placenta revealing developing vertebral column, along with extensive fibrin deposition and foci of calcification in the chorionic villi. (H & E stain, x40)

Discussion

Settegast (1872) is reputed to have been the first to have classically designated the condition “Foetus papyraceus”. The French have called this condition, *un petite bonhomme du pain d’epice* –the little gingerbread man. Pliny (A.D. 23-79) has mentioned about fetus papyraceus in his early writings.^[5] Posner and Klein postulated that “the physical character of the dead fetus would appear to depend upon its position in the uterus with reference to the viable twin, the amount of amniotic fluid in each sac, the integrity of the chorion, and the time of death”. The dead twin is

described as either a “fetus compressus” or “fetus papyraceus”, depending on the degree of flattening.^[6]

Fetus papyraceus cases present two interesting problems. Why should the one fetus die during the early weeks of pregnancy and why should the process of dehydration and compression occur instead of maceration? Browne (1947) attributes the cause of death of one fetus as being due to reversal of the circulation in the hypogastric arteries of the weaker twin by its stronger companion and its placental circulation is thus brought to a standstill. Infarction of that portion of the placenta is usually seen. Velamentous insertion of the cord has been suggested as a possible cause of death.^[5] Marginal insertion of cord has also been postulated, as it was seen in our case.^[1] Other causes include genetic and chromosomal abnormalities, twin-twin transfusion syndrome, true cord knot, cord strictures etc.^[2] Mills (1949) postulated that compression by the sac or the head of the surviving twin causes dehydration with the arrest of maceration and the formation of a fetus papyraceus. Compression may well be a necessary factor, as similar changes in the dead fetus of a single pregnancy have not been reported. Crosman (1936) contended that as the fetal life progresses, enzyme activity becomes more progressive and that in retarded fetuses the reverse obtains. This may be of importance as retarded fetal development may be a precipitating factor, leading to the death and dehydration of the one fetus of a twin pregnancy. Thompson (1927) draws attention to the fact that unless the membranes are intact, the usual process of putrefaction takes place. With intact membranes, the dead fetus is in a sterile medium and so, too, are its own respiratory and alimentary tracts, from which areas putrefaction occurs. Kindred (1944) suggested that the dead fetus is surrounded by a fluid of maternal origin which replaces the liquor amnii and preserves the fetus in a desiccated and embalmed state.^[5]

Fetus papyraceus shows varied morphologic appearances. Fetus papyraceus may be easily apparent at delivery but can appear only as a localised area of thickening within the membranes. Occasionally a fetus papyraceus is sufficiently large to cause obstruction during labour.^[7] Sometimes the dead fetus shrinks and is compressed against the membranes, eventually resembling amorphous necrotic tissue. Undoubtedly many are overlooked.^[8] Sometimes it is embedded within the placental tissue, as it was seen in our case. Similar finding was also reported by Moha-

zab HR in his case.^[9] Hence a careful exploration of the placenta and membranes should be performed as many fetus papyraceus are diagnosed after delivery.^[10]

The primary concern is about the complications to mother and the surviving co twin. Maternal complications include preterm labor, infection from a retained fetus, severe puerperal hemorrhage, consumptive coagulopathy and obstruction of labor by a low lying fetus papyraceus causing dystocia leading to caesarean delivery. In about 90% cases, there is precipitation of pre- term labor and thus risk of prematurity.

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 hydrocephalus, bilateral renal cortical necrosis, unilateral absence of kidney, gastrointestinal tract atresia, gastroschisis, hemifacial microsomia and aplasia cutis. Single fetal death in twin pregnancies exposes mother to the risk of DIC, where breach between maternal and fetal circulation allows passage of tissue thromboplastin from dead fetus and its placenta into maternal circulation leading to hemostasis impairment depending on the intensity of stimulus, incidence being 25%. But the risk of cerebral impairment is the greatest. Anand et al reported that surviving co-twins had poorer scores on the Griffiths Mental and Development Scales when compared to singleton.^[3]

Conclusion

Fetus papyraceus although rare, is a well known entity. It is very important to make a diagnosis in time to prevent severe complications. Thorough inspection of placenta and membranes will permit the diagnosis of abnormalities of its and of fetus papyraceus as occurred in this case.

Acknowledgements

None

Funding

None.

Competing Interests

None declared.

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