Fetus papyraceus—11 cases

E. DAW
F.R.C.O.G.

North Manchester General Hospital, Crumpsall, Manchester M8 6RB

Summary

Eleven cases of the rare condition of fetus papyraceus are described and reviewed.

KEY WORDS: intrauterine death, multiple pregnancy, fetus papyraceus.

Introduction

Fetus papyraceus is a rare condition with the intrauterine death and subsequent retention of one or more fetuses of a multiple gestation. The estimated frequency is 1:12000 live births (0.0085%) (Saier, Burden and Cavanagh, 1975), and the incidence lies between 1:184 and 1:200 twin pregnancies (Saier *et al.*, 1975; Baker and Doering, 1982). It occurs with triplet pregnancy (Skelly *et al.*, 1982) and more multiple sets (Aiken, 1969). This report reviews 11 cases.

Case summaries

(1) A 26-year-old primigravida was found to have a high head at term. Standing lateral pelvimetry showed a fetus papyraceus in the pelvis (Fig. 1). Spontaneous labour followed within 7 days and the fetus papyraceus with its placenta were expelled before the delivery of a healthy female infant 2600 g in weight. The cord of the fetus papyraceus had a velamentous insertion.

(2) This 22-year-old gravida 2 patient was found to have a breech presentation at 38 weeks gestation. A confirmatory abdominal X-ray confirmed not only the breech presentation but also a fetus papyraceus (Fig. 2). At delivery 2 weeks later, a live male infant weighing 2890 g was delivered by the breech with forceps for the aftercoming head. The third stage was uneventful with the fetus papyraceus revealed as a monozygotic twin with no cord complication.

(3) This 28-year-old gravida 3 patient was X-rayed because of uncertain dates. Earlier in pregnancy, her uterine size had been considered large for dates.

The X-ray showed bony parts to one side of the fetus (Fig. 3). Delivery was normal on the expected

FIG. 1. Standing lateral pelvimetry. Fetus papyraceus arrowed.

date estimated by the patient's last menstrual period. The third stage was normal with the fetus papyraceus alongside the placenta and there was no cord complication.

(4) Routine examination of a 26-year-old primigravida's placenta following a normal delivery at 40 weeks revealed a fetus papyraceus (Fig. 4). The monozygotic twin was noted to have a velamentous insertion of the cord.

(5) Routine examination of the placenta and membranes of a 31-year-old gravida 3 patient following delivery revealed a fetus papyraceus. The velam

FIG. 2. Breech presentation. Fetus papyraceus arrowed.

FIG. 3. Fetus papyraceus arrowed.

entous cord insertion appeared to have ruptured and the fetus papyraceus was anaemic in appearance. There was no history of antepartum haemorrhage in the patient's uneventful pregnancy.

(6) A 24-year-old primigravida required a forceps delivery for fetal distress after a spontaneous labour at 38 weeks. The infant weighed 3100 g and required little resuscitation. The third stage was normal but a fetus papyraceus was present with no cord abnormality. However, routine examination of the living infant at the age of 36 hr revealed a systolic murmur and the baby started to have cyanotic attacks. The baby was referred for cardiac investigations, when pulmonary stenosis was diagnosed with a hypoplastic right ventricle. Therapy to maintain the ductus arteriosus was commenced but the infant's condition deteriorated and he died at 5 days of age.

(7) Examination of a 24-year-old primigravida's placenta following a normal delivery at term revealed an 'early' fetus papyraceus (Fig. 5).

(8 and 9) A 25-year-old gravida 5 patient was found to be 3 months pregnant 6 months after her husband had had a vasectomy. X-ray examination at 28 weeks gestation because of polyhydramnios revealed a triplet pregnancy, with one infant showing Spalding's sign. The polyhydramnios disappeared at 32 weeks and the patient delivered one live infant at

FIG. 4. The velamentous cord insertion is arrowed. 37 weeks. Two monozygotic fetus papyraceus were present in the membranes with no cord complications. The surviving infant was normal.

FIG. 5. A rare early fetus papyraceus.

(10) This 24-year-old gravida 3 patient was married to a binovular twin. She was pregnant with an intrauterine contraceptive device *in situ* and had several antepartum haemorrhages which settled with bed rest. She delivered at term a male infant weighing 3210 g. The fetus papyraceus had the cord tightly around its neck.

(11) A gravida 3 patient aged 30 delivered a live infant at 38 weeks. A fetus papyraceus was present in the placenta and membranes.

Discussion

The only sign of the death of one embryo of a multiple pregnancy in the first 6 to 8 weeks may be a cyst on the fetal surface of the surviving infant's placenta. After 8 weeks the death of one embryo in a multiple pregnancy, with resorption of amniotic fluid and mummification of the fetal parts, will cause a fetus papyraceus (Potter, 1962). Death usually occurs during the second trimester and the crown rump length is a good guide to the stage of pregnancy when death occurred, but obviously unless sufficient time has elapsed for mummification, the dead fetus will be delivered as a macerated stillbirth rather than a fetus papyraceus. Therefore delivery of a fetus papyraceus indicates death must have occurred at least 10 weeks previously (Saier *et al.*, 1975).

Antepartum diagnosis of fetus papyraceus is infrequent and usually it is a chance finding during investigation of some other pregnancy problem. The following clinical signs are, however, very suggestive: (i) Rapid enlargement between 12 and 24 weeks gestation, followed by a normal or slowed growth

period and; (ii) the sudden appearance or subsidence of toxæmia of pregnancy. Subsequent delivery of a fetus papyraceus has also explained; (iii) unexplained bouts of vaginal bleeding and (iv) amniotic fluid leakage which suddenly ceases (Saier *et al.*, 1975).

Maternal health is rarely affected, polyhydramnios and toxæmia of pregnancy may suddenly disappear, though complications have been reported—preterm labour, obstructed labour necessitating Caesarean section, infection and postpartum haemorrhage (due to retention of the fetus papyraceus). Consumptive coagulopathy following fetal death in a triplet pregnancy has recently been reported (Skelly *et al.*, 1982).

Velamentous insertion of the cord is increased in twin pregnancy, but rare in fetus papyraceus (Saier *et al.*, 1975). Four of the present 11 cases had velamentous insertion of the cord and another fetus papyraceus had the cord tightly around its neck. This suggests that cord complications may, in fact, increase the chances of intrauterine death and formation of a fetus papyraceus.

No correlation to maternal age, parity or gravidity with fetus papyraceus can be drawn from the 11 cases. Four of the 11 cases were established as monozygotic and one as dizygotic, with the zygosity in doubt in the remaining cases, which suggests that monozygotic multiple pregnancies may be prone to fetus papyraceus formation.

Fetus papyraceus with congenital anomalies in the 2nd twin is rare (Baker and Doering, 1982) though congenital intestinal atresia, gastroschisis, aplasia cutis, congenital and cardiac anomalies have been reported. One of the 11 cases had a cardiac anomaly in the survivor similar to the case of Baker and Doering.

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