Retention of the second twin: a viable option? Case reports

M. G. LONG, D. M. F. GIBB, S. KEMPLEY, L. D. CARDOZO, K. NICOLAIDES, H. GAMSU

Case reports

Patient no. 1

A 38-year-old woman presented in her second pregnancy, her first pregnancy was uncomplicated and ended at term. Twins had been diagnosed in this pregnancy and she was referred at 15 weeks gestation to King's College Hospital for an amniocentesis because of maternal age. An ultrasound scan before the amniocentesis confirmed the gestational age and demonstrated an upper and lower sac. A double puncture technique was used to obtain amniotic fluid samples and the fetal karyotypes were normal. Three days after the amniocentesis she presented to her local hospital with spontaneous rupture of the chorio-amniotic membranes of the lower sac. At 19 weeks gestation she was reviewed at King's College Hospital and a follow-up ultrasound scan confirmed oligohydramnios of the lower sac. All measurements of twin I were below the 5th centile but twin II was growing normally with a normal amount of amniotic fluid. After full discussion with the parents a decision was made to allow the pregnancy to continue. At 21 weeks gestation ultrasound examination revealed persistent oligohydramnios in the lower sac with growth of twin I just

King's College Hospital, Denmark Hill, London SE5 8RX. Department of Obstetrics M. G. LONG D. M. F. GIBB L. D. CARDOZO K. NICOLAIDES Department of Paediatrics S. KEMPLEY H. GAMSU

Correspondence: D. M. F. Gibb.

below the 5th centile. Twin II was still growing normally with normal amniotic fluid volume. The woman was readmitted at 23 weeks gestation with an episode of vaginal bleeding. Two days after admission she developed a pyrexia with tachycardia and antibiotic treatment was started after appropriate swabs had been taken. The uterus became irritable and two days later she complained of a firm mass at the vulva which was found to be a fetal head. Extraction of the fetus was not possible until a general anaesthetic had been administered. The fetus was then gently extracted with the aid of a bivalve speculum, used in the manner of obstetric forceps to the head. The umbilical cord was cut and trimmed at the cervix. Ultrasound examination confirmed an intact second sac with the fetal heart still beating and a conservative approach to further management was adopted. Treatment with antibiotics was continued and nifedipine and ritodrine were prescribed. Over the next 13 days all remained well. At 25 weeks gestation the woman reported an increasing vaginal discharge and an ultrasound scan suggested a slight reduction in amount of amniotic fluid. The vaginal discharge continued and a subsequent scan performed several days later revealed oligohydramnios. At 26 weeks and 5 days gestation, 11 weeks after initial membrane rupture and 21 days after the first twin had been extracted, labour supervened. Shortly after the onset of labour a live female infant was born spontaneously with the aid of an episotomy. The placenta was born by cord traction. Postnatal recovery was uncomplicated. On the second day post partum the placenta of twin I, which had been presumed to have been resorbed, was passed spontaneously. Histological examination of the placentas confirmed the dichorionic, diamniotic nature of the pregnancy with 100% infarction of the smaller placenta.

Neonatal course. The baby weighed 1080 g and had Apgar scores of 5 and 9 at 1 and 5 min respectively. She was electively intubated and intermittent positive pressure (IPPV) was given from birth but she was breathing air by the second day of life and had no further respiratory problems. In view of the prolonged rupture of membranes the baby was given antibiotics for six days after birth but all bacteriological cultures were negative. Oral feeds were started on the third day and she was returned with her mother to the referring hospital one week after the birth. Examination of the infant at 27 weeks of age confirmed that she was healthy and developing normally. Her weight, length and head circumference were all above the 50th centile. She is now 3 years old and well.

Patient no. 2

A 34-year-old woman in her seventh pregnancy had previously given birth to a live girl after induction of labour at 38 weeks gestation for pregnancy-induced hypertension and she subsequently had two first trimester miscarriages, followed by a spontaneous vaginal delivery at 40 weeks gestation and two more first trimester miscarriages. In the current pregnancy she booked at 13 weeks when she gave a history of chronic mild asthma. An ultrasound examination at 15 weeks gestation revealed a twin pregnancy and a further scan at 20 weeks showed that all measurements of both the twins were on the 50th centile. However, a subsequent ultrasound scan at 23 weeks showed no growth in twin I but normal growth in twin II. At this time her asthma worsened due to a chest infection. She was given penicillin and aminophylline and the infection resolved. She presented at 23 weeks and 5 days gestation with vaginal bleeding and she was then admitted to hospital for observation. Three days later twin I was miscarried and the umbilical cord was cut and trimmed at the cervix. Conservative management was continued and the cervix remained closed. Cefuroxime and metronidazole were given for five days. An ultrasound scan at 25 weeks gestation showed that twin II was growing normally but shortly after this rupture of the second sac occurred. An amniocentesis was performed which showed pus cells and scattered cocci and the patient was again given cefuroxime and metronidazole. One day later she became febrile and complained of uterine contractions. Labour was stimulated with oxytocin, followed by the birth of a live girl. The third stage led to uncomplicated delivery of both placentas.

Postnatally the mother continued with a seven day course of the antibiotics and her recovery was uncomplicated.

Neonatal course. The baby weighed 860 g and had Apgar scores of 2 and 7 at 1 and 5 min respectively. She required IPPV for 18 h, then she was extubated and given supplemental oxygen in a headbox. At 3 weeks of age IPPV had to be given again due to recurrent apnoeic attacks associated with bradycardia. By 1 month of age the baby was breathing air. The only other problem was the need for a skin graft of an intravenous infusion site after development of a localized abscess due to *Escherichia coli*. She was discharged home at 68 days of age and at 25 months she is thought to be neuro-developmentally normal. Her height and weight are within the normal range.

Patient no. 3

A 36-year-old woman in her second pregnancy was found to have twins after in-vitro fertilization (IVF). Her first pregnancy had also followed IVF and ended with the delivery at 29 weeks gestation of a severely growth retarded baby weighing 539 g who subsequently died at 2 weeks of age. In the current pregnancy an ultrasound scan at 16 weeks confirmed the twin pregnancy with the biparietal diameter (BPD), head circumference (HC) and abdominal circumference (AC) for twin I on the 45th, 20th and 5th centiles, respectively, and the same measurements for twin II were on the 40th, 20th and 20th centiles, respectively. An ultrasound scan at 21 weeks showed that growth had been maintained, but another scan at 23 weeks and 5 days demonstrated that the AC had dropped to the 5th centile in twin II and below the 5th centile in twin I. Two further ultrasound scans showed that the growth of twin II was maintained but the growth of twin I had slowed markedly and the intrauterine death of twin I occurred at 26 weeks and 5 days gestation. Dexamethasone, 24 mg in divided doses, was administered at 27 weeks and an ultrasound scan at 28 weeks and 5 days showed that the growth of twin II had falled to the 3rd centile. At 29 weeks and 5 days gestation the woman developed abdominal discomfort and shortly afterwards twin I was stillborn and the cord was cut. After discussion with the parents a decision was taken to continue to manage the pregnancy conservatively. Weekly ultrasound scans demonstrated a continued reduction in the rate of growth of the remaining twin. At 32 weeks gestation a live male infant was born by caesarean section. The mother made an uneventful recovery.

Neonatal course. The birthweight was 1100 g and Apgar scores were 8 and 9 at 1 and 5 min. Supplemental inspired oxygen was initially required but the concentration was rapidly reduced and the baby was soon breathing air. Antibiotics were administered initially but were stopped when negative bacteriological cultures were obtained. A brief hypoglycaemic episode was treated with intravenous dextrose and did not recur. Thrombocytopenia was found on day 2 and persisted for the first week. At 10 days he developed abdominal distension and the radiographic appearance was consistent with nonspecific dilatation of the small and large bowel. This resolved after a period of parenteral feeding. Staphylococcus epidermidis septicaemia at 15 days was treated with intravenous vancomycin. He developed conjugated hyperbilirubinaemia and abnormal liver function tests from the age of three weeks. After extensive investigation this was thought to be due to neonatal cholestasis associated with prolonged parenteral nutrition and sepsis. The jaundice gradually resolved and the baby was discharged at 57 days old. When last seen at a chronological age of 48 weeks he was noted to be a tall and thin boy whose neuro-developmental progress was normal.

Patient no. 4

A 31-year-old woman booked at 8 weeks gestation in her second pregnancy. Her previous pregnancy had resulted in a 4·2 kg baby girl born by Neville-Barnes forceps for a prolonged second stage at 41 weeks gestation. She had some vaginal bleeding early in the current pregnancy and an ultrasound scan showed a twin pregnancy. At 17 weeks gestation she presented with a history of ruptured membranes which was confirmed on speculum examination. An ultrasound scan was performed and showed twin I to have BPD, HC, AC and femur length measurements between the 40th and 50th centiles while the same measurements of twin I were on the 50th centile. The sac of twin I showed oligo-

hydramnios and a diagnosis of spontaneous rupture of membranes of this twin was made. Microscopy of a low vaginal swab demonstrated scanty pus cells and broad spectrum antibiotics were administered although culture yielded only normal regional flora. Conservative management was undertaken after discussion with the parents and 2-weekly review was arranged. Ultrasound scans at 19 and 22 weeks gestation revealed progressive reduction in the growth of twin I to <5th centile while the growth of twin II remained constant. The woman then presented at 25 weeks and 4 days with prolapse of the umbilical cord of twin I into the vagina. An ultrasound scan confirmed that twin I had died but also showed that twin II was alive and that its growth had accelerated to between the 60th and 70th centiles. She was given pethidine 75 mg and the umbilical cord, having been pulled further into the vagina, was trimmed as high in the cervix as possible. Antibiotics were prescribed. She was discharged home 24 h later feeling well but she returned the following day complaining of a lump in the vagina and shortly after admission spontaneous expulsion of the first twin occurred. Subsequent examination showed that the cervix had closed with the surviving twin still in utero. Ultrasonography revealed that the remaining twin had reached the 85th centile for gestational age with normal amniotic fluid volume and an estimated fetal weight of 1.2 kg at 25 weeks and 6 days. Conservative management was continued with regular vaginal swabs taken for culture and dexamethasone 24 mg in divided doses was administered after the 26th week. At 27 weeks and 5 days spontaneous rupture of the membranes of the second sac occurred. No contractions were apparent and culture of a low vaginal swab yielded normal regional flora. She remained in hospital for 2 more days when regular contractions started. The presentation was cephalic and vaginal delivery was anticipated. One hour after the contractions had commenced persistent late decelerations of the fetal heart rate occurred. Vaginal examination showed the cervix to be 8 cm dilated with a high presenting part and caesarean section was performed immediately.

The mother's initial recovery after the caesarean section was normal. She continued with the broad spectrum antibiotics but developed pyelonephritis on the second post operative day which responded to gentamicin. She was discharged 7 days after delivery. Neonatal course. The baby, a boy, weighed 1248 g. Apgar scores were 3 and 1 minute and 7 at 10 min. He was given IPPV for 24 h and was then extubated. Antibiotic treatment with penicillin and gentamicin was commenced shortly after birth. Surface cultures grew E. Coli but blood cultures were negative. An unconjugated jaundice was treated with phototherapy. At 6 days of age, having started enteral feeding, he developed necrotizing enterocolitis which responded to antibiotics and parenteral nutrition. At 37 days of age he developed bronchiolitis due to respiratory syncytial virus and required IPPV and treatment with nebulized Ribavarin. Over the next few days additional inspired oxygen was gradually reduced until he was breathing air.

He was discharged home at 2 months of age and follow-up until the most recent visit at 14 months of age has not revealed any neurodevelopmental problem. Subsequent admissions have, however, been necessary for the treatment of pneumonia, wheezing and a single febrile convulsion. In addition, he has had recurrent otitis media which has necessitated the insertion of grommets. It has now been established that he has a serum dependent inherited defect of complement which affects phagocytosis and killing by leukocytes. This will require long-term antibiotic therapy in childhood. His weight and head circumference are in the normal range.

Discussion

The complications of twin pregnancy and their contribution to early pregnancy loss and perinatal mortality rates are well known. Spontaneous rupture of the membranes of one sac followed by the birth of the first twin usually results in the pregnancy being terminated because of the traditional belief that a poor outcome is certain. A successful outcome for a remaining twin is, however, not uncommonly seen following the death of one twin in utero without membrane rupture. Death may occur early, resulting in a fetus papyraceous, or later in the pregnancy resulting in the birth of the dead twin with its live counterpart. Continuation of the pregnancy after one twin has miscarried is rare. Four cases are presented in this report where conservative management was adopted

after early spontaneous rupture of the membranes of the first sac and subsequent expulsion of the presenting fetus has occurred. In each case the pregnancy continued and the second twin survived with no discernible long-term problems that could be ascribed to the unusual circumstances which occurred in the pregnancy.

Williams & Cummings (1953) reported a twin pregnancy with one fetus in each horn of a uterus didelphys where the second twin was born 57 days after labour had resulted in the birth of the first twin at 32 weeks gestation. A similar case was reported by Dorgan & Clarke (1959) where the birth interval between twins was 21 days. Reports describing prolonged intervals between the births of twin fetuses in a normal uterus are rare. Abrams (1957) described a case where the first twin miscarried at 23 weeks gestation and the second twin was born 5 weeks later and survived. Druker et al. (1960), reported a case in which a fetus miscarried in an undiagnosed twin pregnancy after 18 weeks gestation and the presence of the second fetus was revealed after it was noticed that the uterus remained larger than would have been expected from a retained placenta. The second fetus was then born after spontaneous labour 65 days later. A more recent case of a first twin miscarrying at 21 weeks gestation, followed by the successful birth of the second twin at 33 weeks gestation was described by Feichtinger et al. (1989).

Tacchi & Dunlop (1987) reported prolapse of the umbilical cord of one of a set of triplets at 19 weeks gestation. After treatment with ritodrine and antibiotics the leading fetus was extracted under anaesthesia, the cord was excised close to the placental disc and a mersilene tape cervical suture was inserted. Surviving twins were born 9 weeks later.

The present report appears to be the largest series in which conservative management of twin pregnancy was employed after the first fetus had been expelled from the uterus. The time interval between the births of the fetuses ranged from 8 to 21 days and in each case there had been severe growth impairment of the first twin. In three of our patients this followed rupture of the membranes and development of oligohydramnios. Prophylactic antibiotics were prescribed after rupture of the membranes of the first twin to discourage the development of chorio-amnionitis which would have jeopardized the survival of the second twin.

Although the birthweight of one baby (patient

no. 3) was <3rd centile, the other three had birthweights >50th centile for their gestational age. Growth of all the surviving fetuses had been noted to be better than that of their co-twins who subsequently died. In patient 4, death of the discordantly small fetus was followed by a growth spurt in the survivor. We postulate that uterine blood flow, already increased in pregnancy, is even greater in twin pregnancy to accommodate the metabolic needs of two babies. When one twin dies, then the augmented blood flow will be available to the survivor who would consequently gain more weight than expected in the period before delivery.

A factor of great importance in the survival of the remaining twin is the availability of a neonatal intensive care unit: all the babies in our series needed specialist support. It is encouraging that all of them are neuro-developmentally normal at the time of reporting with follow-up periods ranging from 11 months to 3 years.

References

- Abrams R. H. (1957) Double pregnancy: Report of a case with 35 days between deliveries. Obstet Gynecol 9, 435–438.
- Dorgan L. T. & Clarke P. E. (1956) Uterus didelphys with double pregnancy. Am J Obstet Gynecol 72, 663–666.
- Drucker P., Finkel J. & Savel L. E. (1960) Sixty-five day interval between the births of twins. A case report. Am J Obstet Gynecol 80, 761–763.
- Feichtinger W., Breitenccker G. & Frohlich H. (1989) Prolongation of pregnancy and survival of twin B after loss of twin A at 21 weeks gestation. Am J Obstet Gynecol 161, 891-893.
- Tacchi D. & Dunlop W. (1987) A multiple pregnancy after in vitro fertilization. Case Report. Br J Obstet Gynaecol 94, 1223–1224.
- Williams B. & Cummings G. (1953) An unusual case of twins. A case report. J Obstet Gynaecol Brit Emp 60, 319–321.

Received 15 April 1991 Accepted 27 June 1991