

Acknowledgements

The author wishes to thank Dr Emyr J. Jones, consultant physician and Dr Hubert Jones, consultant radiologist and their staffs at the C. & A. General Hospital, Bangor, for their co-operation, also Dr T. F. Jarman, Mass Radiography Service, Welsh Regional Hospital Board and Dr W. E. Miall, Pneumoconiosis Research Unit, Llandough Hospital, Penarth, Glamorgan for their help.

REFERENCES

- Caplan, A. (1953). *Thorax*, **8**, 29.
Cough, J., Rivers, D. and Seal, R. M. E. (1955). *Thorax*, **10**, 9.

A CURIOUS TWIN ABORTION

N. E. MELLING, M.B., CH.B.

Sennybridge

THE FOLLOWING CASE IS UNIQUE in my experience, and, as it raises several problems in genetics and embryonic development, I felt that it might be worth submitting for publication.

Case History. My patient Mrs B. is a housewife, 26 years of age. She is one of a family of eight. Her husband G. is 35, and one of a family of six. There is no history of twinning in either family, but B's aunt by marriage is a twin, and one of her daughters, a woman of about B's age, has recently given birth to twins. B. and G. have two children, both girls, aged 6 and 4 years respectively, and I attended Mrs B. throughout both pregnancies, labours and post-natal periods. They were normal in all respects, and there has never been any suggestion that her uterus might be bifid. After the birth of the baby in 1958 the couple decided that they did not want any more children, and have used an occlusive cap, with "Volpar" paste or gels consistently since. Supplies ran out in July, 1962, however, and an unprotected coitus took place on the 20th of that month. B. came to see me on 3 August, fearing that she might be pregnant. Her last menstrual loss had started on 29 June. She has a regular 28-day cycle, so the next period was due on 27 July, but this had not shown up. We had some discussion as to whether the coitus of 20 July was within the safe period or not. Presumably she had ovulated at about 13 July, i.e. 14 days after 29 June, so, as far as we could see at the time, she was probably "safe". (Malleon and Blacker, 1950). To make certain, I gave her a course of "Amenorome" tablets as a pregnancy test. Each tablet contains ethinyloestradiol 0.01 mg. and ethisterone 10 mg. and my usual habit is to give four tablets daily for five days. In the absence of a pregnancy, withdrawal bleeding may be expected within the week following the end of the course. In addition, because of her anxiety I gave her 1 gr. "Spansule" of phenobarbitone each morning.

She came to see me again on 3 September. The pregnancy test had been positive—i.e. there had been no withdrawal bleeding, and, although they had not really wanted to increase their family, both B. and G. had accepted the fact philosophically. They had discarded their contraceptives and had continued intercourse as they had always done. There was at least one coitus during the family holiday, from 17 to 30 August, and perhaps two or three more from then until the events of 14 September—now that they knew the worst, they reverted to the vagueness about dates that is normal in this field of human relationships.

At this antenatal examination (3 September) I thought that I could just feel the womb behind the symphysis pubis, this indicating that it was larger than was to be expected, for I do not reckon to be able to feel it abdominally with any confidence before the twelfth week. B. was booked for a home confinement, and asked to come again in a month's time.

On 12 September she was involved in an accident on one of our narrow country lanes. By skilful driving, she managed to avoid a collision with another vehicle, but took her car over a steep bank in the process. She was severely shaken but otherwise apparently unhurt and did not consult me about it. Two days later, she was awakened at 5.30 a.m. by severe lower abdominal pain; at 7 a.m. she started to bleed quite briskly, and an hour later sent for me. At 8.30 a.m. I found her womb enlarged to four fingers' breadths above the symphysis pubis, hard and tender. The os admitted the tip of one finger, and a steady trickle of blood was coming from it. I gave her "Ergorondase" (ergometrine maleate 0.5 mg. with hyaluronidase 330 IU) with "Hyperduric" morphine gr. $\frac{1}{4}$ and 50 mg. "Sparine", all intramuscularly. By 9.0 a.m. she was more comfortable and drowsy, and I left. Shortly after this, the two embryos described below were discharged. At my next visit at noon, I found the womb was still enlarged, but not so tender as formerly, and I put her on "Femergin" (tablets of ergometrine tartrate, 0.001 G.) one every six hours. That evening she passed a piece of decidua, about 5 x 3 x 1 cm., and the womb contracted into the pelvis. Further progress was uneventful. There was no sign of lactation throughout the postabortum period, and at the final examination on 5 October, I found nothing abnormal in the pelvis.

The Embryos

The two embryos are as illustrated in the photograph. I was unable to identify any other products of conception in the blood clot and detritus that had come away during the miscarriage, and so cannot say whether the twins were monovular or binovular. The smaller embryo is 16 mm. in length, and weighs 0.6 G. (after fixation in formalin). The eye spots, auriculae and limb buds are present, but I cannot identify any digits. Rudimentary and undifferentiated sex organs are present. This embryo is apparently normal. The larger individual is a bad one. Its total length is 28 mm. and its weight (again after fixation) 2.5 G. Of the length, 12 mm. are accounted for by the legs. These are fused as far as the insteps, the normal division being represented by a shallow anterior groove. The remaining 16 mm. do not represent the full crown-rump length, for the embryo is anencephalic. The mouth is wide, and the face protrudes as a well-marked snout. The arms, hands and fingers are well formed, and flexed across the chest—a position that, with the facial deformity, makes the whole look like a pugnacious amphibian! On the front of the lower abdomen is a membranous protruberance, presumably the normal umbilical hernia that is present at this stage of development. The sex organs are represented merely by a circular plaque, 2mm. in diameter.

Estimation of the age of an embryo is fraught with difficulty, even for the professional: for the amateur, it becomes a matter of more or less (un)inspired guesswork! It seems to me that the smaller one

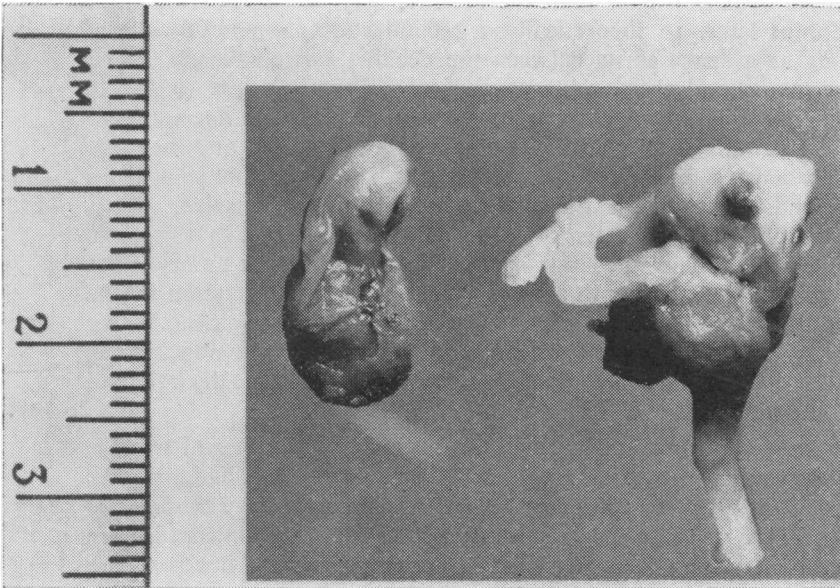


FIG. 1
A Curious Twin Abortion

of these two represents at least six to seven weeks gestation, and the larger one, allowing for its deformities, at most seven to eight weeks, and these figures would fit in with the known length of the pregnancy, counting from 20 July, and not from the last menstrual period. The whole of embryonic and early foetal life is, however, a time of extremely rapid growth. In six weeks, the fertilized ovum measuring about 140μ in diameter, has become 16 mm. long: in the next six weeks, it will increase in length to 10 cm. Thus it is possible that the difference between these two embryos is of the order of two or three days only.

Discussion

The most striking feature in this case is the disparity in the stages of development reached by the two embryos in a single uterus. Such a difference can arise in one of two ways: either (a) two separate conceptions have taken place at different times (superfoetation or superfecundation) i.e. the difference in age is real: or (b) the twins are conceived at the same time, but grow at different rates, i.e. the difference is only apparent.

Superfoetation means the superimposition of a new pregnancy on an already existing one, and so implies an interval between the two of at least one month—the second pregnancy must be at least one

whole ovulation later than the first. It may be two or even three months later, as, theoretically, a second conception is possible as long as a space exists between the decidua capsularis of the first pregnancy and the decidua vera (Kenny, 1951; Gaupin, 1951). In superfecundation, two ova are released together, as in normal binovular twinning. One is fertilized following one coitus, and the other after a later one—and the time interval at which this is possible depends on the time that the second ovum can survive, and is probably only a matter of hours (Lennon, 1962). In this case, superfoetation is ruled out by the fact that the two embryos are much less than a month apart in age, and superfecundation by the reliable history of only one coitus on 20 July. In any case, if ever the question of additional conceptions arises in human beings, some other and simpler explanation of the facts can usually be found (Lennon, 1962).

We were taught as students that inequality in the size of twins is to be explained by assuming that the smaller one has had a poorer blood supply than the other (the Queen Charlotte's *Text-book of Obstetrics*, 1936). Professor McCance, in his first Lumleian lecture for 1962, accepts this possibility in later pregnancy, but is of the opinion that, regarding general mammalian development *in utero*, the weight gain in all embryos in a litter is the same at first, and does not depend on the genetic background, the number or the position of any individual in the uterus (McCance, 1962). My case contradicts this: for of the two embryos in this "litter", one is four times as heavy as the other. It seems to me that this must be regarded as a case of normal twinning, in which one embryo, a bad one as it happens, has had, for some unknown reason, a priority claim on the uterine blood supply.

The second interesting feature is the occurrence of anencephaly in one of the twins. It is usually held that anencephaly is due to a genetic defect, a Mendelian recessive (Boyd and Hamilton, 1960) in which the neural tube fails to close between the third and fourth weeks and the consequent exposure of the neural plate to the amniotic fluid causes it to atrophy. Thus Lewis (1960) reports a case of binovular twin pregnancy in which both fetuses were anencephalic—one being a male, which is unusual. He refers to four cases that show a similar double dose of genetic bad luck, and also to a report of seven cases of twins in each of which only one foetus was affected. He does not say whether these were cases of uniovular or binovular twinning, but Pedlow (1961) described a case of mono-amniotic twins only one of which showed the anomaly. These twins were females, as the majority of anencephalics are: they were born at 41 weeks, the abnormal one being stillborn, but her sister being healthy and living. The same report refers to two similar cases in

the literature, and one of di-amniotic monochorionic twins, where only one was affected. Such cases are parallel to mine, although I, regrettably, am unable to say whether my twins were uniovular or binovular. They throw doubt on the orthodox view: for uniovular twins must have the same genetic make-up, and, in anomalies arising from genetic defect should both be affected. Their occurrence poses the question as to whether, pathogenically, there are at least two types of anencephaly.

Finally, what brought about the miscarriage in this case? Four possible factors present themselves:

(a) The malformation of one embryo, causing intra-uterine death, and consequent expulsion of what was then a foreign body. Anencephaly, however, is not necessarily a cause of abortion. We all know to our cost that such monsters can go to term—I have delivered four in this practice since 1945.

(b) The possible abortifacient action of the drugs used early in the pregnancy. We are always assured that hormone preparations designed as pregnancy tests—there are several of them—are safe; and phenobarbitone has a long history and an honoured name. The recent thalidomide tragedy has forced us to look with suspicion upon all medicaments used when the embryo is starting its development—though admittedly we are more concerned with possible teratogenic effects than with abortifacient ones. In the light of this scrutiny, are our old friends as benign as we have believed?

(c) The lateness of the impregnating coitus in the menstrual cycle. There is a strong presumption that B. ovulated on or about 13 July, but no positive proof: for regularity in the menstrual cycle is always a relative term—the best of women vary. If ovulation did occur on the expected date, the ovum (or ova) was at least seven days old by the time it was fertilized (since coitus and conception are not synchronous). This brings it within the stale-egg concept (Roth, 1962) which argues that late fertilization of a healthy ovum that has begun to degenerate may be the cause of casual spontaneous abortion.

(d) The motor accident of 12 September. This is probably the villain of the piece—but we all know women who have been shaken much more severely, and even badly injured, without their pregnancies being affected. It may thus be that the accident was only the precipitating cause, the pregnancy having been unstable from the beginning as the result of one or more of the other factors.

There is one small point to make in conclusion. The time-honoured formula for calculating the expected date of delivery from the first day of the last menstrual period would have given too early a date in this case. We have, most unusually, a firm date for the impregnating coitus, and we know that it came late in the cycle, and this would give us a much more satisfactory base line to start from. The absence of such information in the vast majority of pregnancies is one of the reasons why estimating when a baby will arrive is more a sport than a science.

Summary

A twin abortion at eight weeks, one of the embryos being anencephalic and at a later stage of development than the other, is described. The problems of the disparity in growth, of the presence

of anencephaly in one of the twins, and of the cause of the miscarriage are discussed.

Acknowledgements

I am indebted to my patients for their ready co-operation, and their permission to publish their case: to the department of zoology in the National Museum of Wales for the photograph: and to Professor G. G. Lennon, of the department of obstetrics and gynaecology in the University of Bristol, for his encouragement and advice.

REFERENCES

- Boyd, J. D., and Hamilton, W. J. (1950). *Modern Trends in Obstetrics and Gynaecology* (Butterworth) Chapter 11.
- Gaupin, C. E. (1951). *Amer. J. Obstet. Gynec.*, **62**, 212 (quoted in *Lancet Annotation*, 1953, **1**, 478).
- Kenny, M. (1951). *British Encyclopaedia of Medical Practice* (Butterworth). Vol. 7, p. 501.
- Lennon, G. G. (1962). *Diagnosis in Clinical Obstetrics* (Wright) p. 163.
- Lewis, L. (1960). *Brit. med. J.*, **2**, 1500.
- Malleson, J. and Blacker, C. P. (1950). *British Encyclopaedia of Medical Practice* (Butterworth). Vol. 3, p. 625.
- McCance, R. A. (1962). *Lancet*, **2**, 621.
- Pedlow, P. R. B. (1961). *Brit. med. J.*, **2**, 997.
- Queen Charlotte's *Textbook of Obstetrics* (1936). 4th Edition. Chapter XVI.
- Roth, D. B. (1962). *Obstet. and Gynaec.*, **19**, 411.
-

NEWS FROM ABROAD

REPORT ON THE FIFTH INTERNATIONAL CONGRESS FOR GENERAL PRACTICE

held in Salzburg from 15 to 22 September 1963

THIS CONGRESS WAS ATTENDED by about 150 practitioners, seven-eighths from Austria and Germany, the remainder from Switzerland, Hungary, Denmark, Sweden, Belgium, Canada, U.S.A., Australia and the United Kingdom.

The programme was varied, having several excellent papers, such as on (1) the part played by psychology in general medicine, (2) the scope and educational need of the general practitioner, (3) head injuries, (4) early diagnosis.

Six to eight papers were read and discussed each day. Here the organization of the Congress was not strict enough as almost every speaker over-