

Errors in Birth Registrations and Coding of Twins and Higher Order Multiples

Peter O.D. Pharoah

FSID Unit of Perinatal and Paediatric Epidemiology, Department of Public Health, University of Liverpool, United Kingdom

Dizygotic compared with monozygotic conceptions are at decreased risk of fetal and infant death and serious morbidity in surviving infants. Different sex twin maternities must be dizygotic but miscoding and incorrect registration of sex and number of fetuses may lead to an incorrect assignment of zygosity. The aim of the study was to validate the coding and registration of number and sex of births in multiple pregnancies. Fetal and infant death registrations from all multiple maternities in England and Wales 1993–1998 were examined. There were 51,792 twin, 1627 triplet and 51 higher order multiple maternities that were registered. Among these there were 1926 fetal deaths, 58 of which were registered as being of indeterminate sex but were coded as male in 56 and female in 2 cases. A fetus *papyraceous* was registered as male in 19 and as female in 19 cases. Other fetal deaths weighing $\leq 100\text{g}$, with no mention of *papyraceous* on the death certificate, nevertheless, likely to be of indeterminate sex, were registered as male in 26 and as female in 23 cases. In 13 maternities, the number of infants registered at birth was less than the number mentioned on the registration certificate. It cannot be assumed that multiple births of different registered sex are dizygotic. As surviving infants from a monozygotic multiple birth are at much greater risk of infant death and serious morbidity than dizygotic multiple births, incorrect assignment of sex has important implications for parental counselling and may have medico-legal relevance when attributing negligence as the cause of morbidity in a survivor from a multiple pregnancy.

A fetal death is defined as:

Death prior to the complete expulsion or extraction from its mother of a product of reproduction, irrespective of the duration of the pregnancy; the death is indicated by the fact that after such separation the fetus does not breathe or show any other evidence of life, such as beating of the heart, pulsation of the umbilical cord, or definite movement of the voluntary muscles (World Health Organisation, 1977).

The registration of a fetal death is a legal requirement and, in the United Kingdom since 1992, a completed gestational age of 24 weeks separates a fetal death from a spontaneous abortion. Multiple births have higher fetal death rates than singleton births (Campbell & MacGillivray, 1988; Kiely, 1990) and, within multiple births, monozygous (MZ) have higher rates of fetal and infant death than dizygous (DZ) conceptions (Myriantopoulos, 1970; Potter, 1963).

National statistics, compiled from birth registrations, do not allow a distinction between MZ and DZ conceptions. However, specifically for twins, national data are published according to whether they are of the same or of

different sex. This allows a partial separation because all different sex twins are DZ but same sex twins comprise both MZ and DZ twins.

Because zygosity plays such an important role in fetal and infant mortality and morbidity, national statistics on multiple births were examined to assess their completeness of ascertainment and their validity in differentiating between same and different sex multiple births.

Methods

The Office for National Statistics (ONS) provided the non-confidential sections of fetal death, livebirth and infant death registrations of all multiple births in England and Wales for 1993 to 1998. This allowed an examination of the sex registered at birth, the coding of the registered sex, the certified cause of death and the plurality of birth, that is, whether registered as twins, triplets, quadruplets or higher order.

In certifying a fetal death, the certifier is not required to state if it is a fetus *papyraceous*, nevertheless, this description frequently appears among the causes of death. Search was made for any mention of fetus *papyraceous*, fetus *compressus* or mummified fetus among the causes of death and it was assumed that such fetuses were of indeterminate sex.

Also, it is not necessary for the certifier to indicate the plurality of the pregnancy, nevertheless, this sometimes appears among the causes of death. For all death certificates from registered twin births, search was made for any mention of triplet or higher order pregnancy among the causes of death.

As it is probable that the sex of fetal deaths of extremely low birthweight would be indeterminate on routine clinical examination, search was made for all fetal deaths of birthweight less than or equal to 100g to determine what sex had been registered.

Results

Errors in Coding of Registered Sex

There were 103,584 twin births in 1993–8 of which 1793 were registered as fetal deaths, a stillbirth rate of 17.3 per 1000 total births. Of the 1793 fetal deaths, 40 (2.2%) were

Address for correspondence: Peter O.D. Pharoah, FSID Unit of Perinatal and Paediatric Epidemiology, Department of Public Health, University of Liverpool, Liverpool L69 3GB, United Kingdom. Email: p.o.d.pharoah@liv.ac.uk

registered as being of “indeterminate” sex. However, for the purpose of national statistics, 38 of the 40 indeterminate sex were coded as male and 2 as female. The 2 coded as female were both born in the latter part of 1998.

There were 4881 triplet births of which 122 were registered as fetal deaths, a stillbirth rate of 25.0 per 1000 total births. Of the 122 fetal deaths, 11 (9.0%) were registered as being of indeterminate sex and all were coded as male.

There were 209 quadruplet or higher order births with 11 registered fetal deaths, a stillbirth rate of 52.6 per 1000 total births. Of the 11 fetal deaths, 7 (63.6%) were registered indeterminate sex and all were coded male.

Two livebirths, both very preterm weighing less than 1500g and dying within an hour of birth were registered as indeterminate sex but coded as male.

The number of fetal deaths registered as indeterminate sex by year and multiplicity of birth is shown in Table 1.

Errors in the Sex That is Registered at Birth

The onus for registering a birth is with the parents. If a birth is of indeterminate sex, the registrar of births has the discretion to allow the parents the choice of what sex is registered.

The 133 fetal deaths among the triplet and quadruplet pregnancies included four that were certified as fetus papyraceous. All four were registered as being of indeterminate sex. An additional 11 registered fetal deaths weighed ≤ 100g of which two were registered as male and seven as female. Assuming that the sex of these 11 extremely small fetal deaths could not be accurately assigned on routine clinical examination, it may be inferred that the registration of the sex was potentially incorrect in 8%.

Among the 1793 registered fetal deaths from twin pregnancies, 55 were certified as fetus papyraceous. Of these, 17 were registered as indeterminate sex, 19 were registered as male and 19 as female. Therefore, in 69% of cases where the sex of the fetal death can be assumed to be indeterminate, the parents were allowed to exercise choice in the sex that was registered. An additional 49 fetal deaths were of birthweight ≤ 100g, 26 were registered as male and 23 as female. Thus, among fetal deaths in twin pregnancies, for 87 (4.9%) the registered sex was potentially incorrect.

It is not known which parents exercised choice in the sex that was registered or whether the sex chosen to be registered was influenced by the sex of the surviving twin.

There were 55 instances where fetus papyraceous appeared as a cause of death on the fetal death certificate. This was noted among 103,584 twin births, a prevalence of about 1:2000 twin births (or 1:1000 twin maternities).

Errors in the Number of Births Registered in Multiple Pregnancies

Among the fetal and infant death registrations, there were two instances where it was stated that the pregnancy was quadruplet but only triplet births were registered. In a further two cases of quadruplet pregnancy only twins were registered at birth.

Similarly, there were nine cases among the fetal and infant death registrations where a triplet pregnancy was noted but only twin births were registered.

Discussion

Before 1998, the guidance given to coding clerks was that all births of indeterminate sex should be coded as male. Thus the coding of a fetal death, and even the occasional very preterm livebirth, will affect the relative numbers of same and different sex twins published in the national statistics. Late in 1998, the guidance given to the coding clerks was changed and births of indeterminate sex were altered to alternate coding of male and female. Unfortunately this change will not remedy the deficiency. Fetuses of indeterminate sex need to be coded as such and dealt with as a separate entity. Weinberg’s rule uses the number of same and different sex twins to estimate the number of MZ and DZ twins (Weinberg, 1902) and, after application of Weinberg’s rule to national data, the relative risk of mono- compared with dizygous twins for fetal death has been determined (West et al., 1999). Coding error in the sex registered means that about 50% of twins with a fetal death of indeterminate sex will be designated as being of the same sex and 50% of different sex. Because MZ are at greater risk of fetal death than DZ conceptions, the relative risk will have been underestimated by such coding error of the sex of a fetal death.

In an analysis of national data, allowance can be made for these coding errors by examining the original death certificate and excluding those registered as indeterminate sex. However, the discretion exercised by the Registrars of Births in allowing parental choice in what sex is registered when the fetal death is of indeterminate sex, cannot be rectified by an examination of the death certificate. This has potentially serious implications. In a MZ multiple pregnancy, death of one fetus is associated with a high risk of death or serious morbidity in the co-conceptus (Pharoah & Adi, 2000). When a fetal death and a co-twin survivor have been registered as being of different sex, it cannot be assumed that the conception was DZ and counselling of the parents about the risk of serious morbidity in the surviving twin must take this into account. It is understandable that parents wish to mourn the death of a fetus and therefore, to assign a sex to the fetus that died. Nevertheless, it should be possible for the Registrar of Births to register a specific sex but to annotate it with a symbol that will enable recognition that the sex was not determinable at birth.

Table 1
Annual Number of Multiple Births Registered as Being of “Indeterminate” Sex

Year	Twins	Triplets	Quadruplets or more	Total
1993	4	0	0	4
1994	6	0	2	8
1995	6	3	0	9
1996	3	2	0	5
1997	12	3	3	18
1998	9	3	2	14
1993–8	40	11	7	58

Failure to register one or more conceptuses from a multiple pregnancy also has implications for counselling parents about the prognosis of surviving livebirths. When the sex of a fetal death is incorrectly registered, it should not be assumed that twins of different sex were both from a DZ conception. Failure to register one or more conceptuses in a multiple pregnancy may occur because a fetus that died has been expelled before 24 completed weeks of gestation and there was no legal requirement for registration. This is liable to occur in triplet and higher order pregnancies that are very preterm at delivery or where there has been selective fetocide before 24 weeks gestation. Alternatively, although registration was legally required because expulsion of the dead fetus occurred after 24 completed weeks of gestation, the parents may not have been informed that a twin had died in utero and did not register the fetal death. This is most likely to occur with a fetus papyraceous (Heys, 1996; Pharoah & Cooke, 1997a). Omitting to inform the parents may be deliberate in order not to cause them distress.

Failure to register a fetus papyraceous from a twin pregnancy has particular relevance if the surviving co-twin suffers serious morbidity such as cerebral palsy or severe learning disability. In these cases, the surviving twin is registered as a singleton and the possibility that the cerebral impairment is a consequence of twinning is not considered. Although the cause of the cerebral impairment may not influence significantly the subsequent management of the child, it does have serious implications for medical litigation. It is not possible, from national statistics, to determine how frequently a fetus papyraceous is not registered. In the data reported here, the prevalence of a fetus papyraceous was about 1:2000 twin registrations which is considerably less than the 1:200 prevalence reported elsewhere (Ottolenghi-Preti, 1972), suggesting there may be a significant degree of under-registration. Among 12 cases of fetus papyraceous reported to the Confidential Enquiry into Stillbirths and Deaths in Infancy in Wessex, only five were registered in the national statistics (Gompels & Davies, 1999). The failure to register one or more fetal deaths in a multiple pregnancy led to the hypothesis that cerebral palsy in an apparently singleton infant may be the consequence of very early fetal loss as the "vanishing" twin (Pharoah & Cooke, 1997b).

Litigation for medical negligence in cases of cerebral palsy is escalating. It is becoming even more imperative that every effort is made to record, as accurately as possible, the degree of multiplicity of a pregnancy, any fetal loss irrespective of the gestational age of that loss, the gender of the lost fetus and the type of placentation.

An infant with pre-partum acquired cerebral impairment as a result of twinning may be more prone to fetal distress labour and to require resuscitation. The cerebral

impairment may then be incorrectly attributed to peripartum clinical negligence.

Acknowledgments

The cooperation and patience of Beverly Botting, Nirupa Dattani and Steve Rowan of the Office for National Statistics in providing the registration data and in answering innumerable queries is gratefully acknowledged. The author is also grateful for funding by the Foundation for the Study of Infant Death (FSID)

References

- Campbell, D. M., & MacGillivray, I. (1988). Outcome of twin pregnancies. In I. MacGillivray, D. M. Campbell, & B. Thompson, *Twinning and twins* (pp. 179–205). Brisbane: John Wiley Ltd.
- Gompels, M. J., & Davies, D. (1999). Fetus papyraceous is being registered increasingly in Wessex. *British Medical Journal*, *319*, 1271.
- Heys, R. F. (1996). Selective abortion. *British Medical Journal*, *313*, 1004.
- Kiely, J. L. (1990). The epidemiology of perinatal mortality in multiple births. *Bulletin of the New York Academy of Medicine*, *66*, 618–637.
- Myriantopoulos, M. C. (1970). An epidemiologic survey of twins in a large prospectively studied population. *American Journal of Human Genetics*, *22*, 611–629.
- Ottolenghi-Preti, G. F. (1972). Sopra un rarissimo caso di gravidanza gemellare con un feto papiraceo e con inserzione velamentosa del funicolo del feto vivo. *Annali Ostetrica, Ginecologia, Medicina perinatale*, *93*, 173–199.
- Pharoah, P. O. D., & Adi, Y. (2000). The consequences of fetal death in a twin pregnancy. *Lancet*, *355*, 1597–1602.
- Pharoah, P. O. D., & Cooke, R. W. I. (1997a). Registering a fetus papyraceous. *British Medical Journal*, *314*, 441–442.
- Pharoah, P. O. D., & Cooke, R. W. I. (1997b). A hypothesis for the aetiology of spastic cerebral palsy — The vanishing twin. *Developmental Medicine and Child Neurology*, *39*, 292–296.
- Potter, E. L. (1963). Twin zygosity and placental form in relation to the outcome of the pregnancy. *American Journal of Obstetrics and Gynecology*, *87*, 566–577.
- Weinberg, W. (1902). Beiträge zur Physiologie und Pathologie der Mehrlingsgeburten beim Menschen. *Archiv für die gesamte Physiologie des Menschen und der Tiere*, *88*, 346–430.
- West, C. R., Adi, Y., & Pharoah, P. O. D. (1999). Fetal and infant death in mono- and dizygotic twins in England and Wales 1982–91. *Archives of Disease in Childhood*, *80*, F217–F220.
- World Health Organisation (1977). *Manual of the international statistical classification of diseases, injuries, and causes of death* (Vol. 1; 9th rev.). Geneva: World Health Organisation.