



Acta Genet Med Gemellol 37:339-345 (1988)
©1988 by The Mendel Institute, Rome

Received 28 June 1988
Final 22 September 1988

Conjoined Twins in Japan, 1979-1985

Yoko Imaizumi

Institute of Population Problems, Ministry of Health and Welfare, Tokyo, Japan

Abstract. Nation-wide data in Japan on the 112 sets of conjoined twins from fetal deaths and from postnatal deaths during 1979-1985 were analysed. Female conjoined twins accounted for 60% of cases. The incidence rate of conjoined twins remained constant except in 1985. Overall incidence rate was 10 per million births. Maternal age effect was found in mothers over the age of 40, where the highest incidence rate was obtained. The incidence rate of conjoined twins increased with birth order. There was no seasonal variation in the time of conception.

Key words: Conjoined twins, Maternal age, Birth order, Geographical variation, Seasonality.

INTRODUCTION

The cause of conjoined twins is unknown. Bhattay et al [2] mentioned that an epidemic of conjoined twins in Southern Africa may be the result of environmental factors.

Recently, Zake [7] reported a higher incidence of conjoined twins in Uganda, the rate being 1:4,242 deliveries. The incidence ranged between 1:2,800 in India and 1:200,000 births in the United States [3]. Edmonds and Layde [3] suggest that a better estimate is between 1:100,000 and 1:30,000 births.

The present study examines secular change and seasonal, geographical and social class variations in the incidence of conjoined twins during the period from 1979 to 1985. It also deals with the effect of maternal age and birth order on this incidence.

MATERIALS AND METHODS

Two sets of data were used. The first, comprising 96 fetal deaths from conjoined

twins reported during the period from 1979 to 1985, were obtained from the fetal death certificate records kept by the Ministry of Health and Welfare. In Japan, data on fetal deaths are available only after the beginning of the 13th week of fetal life. The certificate includes information on parental ages, the occupation of the head of household, sex of the fetus, numbers of previous pregnancies and live births, period of gestation, place and date of fetal death, and cause of death.

The second, comprising data on 26 postnatal deaths from conjoined twins reported during the period from 1979 to 1985, were taken from the death certificate records kept by the Ministry of Health and Welfare. The death certificate records give information concerning the occupation of the head of household, sex, current address, dates of birth and death, and cause of death. Among 26 postnatal deaths from conjoined twins, 20 dead twin persons were classified into ten sets of twin pairs, but cotwins for six dead persons from conjoined twins were not identified from the death certificate record. Information on maternal age, birth order, and gestational age for ten sets of conjoined twins were obtained from the birth certificate record. Data on the general population were taken from the annual volumes of "Vital Statistics of Japan for the Years 1979-1985" (Health and Welfare Statistics and Information Department, Ministry of Health and Welfare), which covers the entire population of Japan. In computing the incidence of conjoined twins, the number of twin sets of pre- and postnatal deaths from conjoined twins were divided by the total number of births (the number of all fetal deaths over 12 weeks plus live births) in the general population during the same period.

Table 1 - Conjoined twin sets, 1979-1985

Year	Fetal deaths				Postnatal deaths					Grand total	Incidence (conjoined twins per million births)
	Sex of twin sets				Sex of twin sets						
	MM	FF	??	Total	MM	FF	M	F	Total		
1979	10	9	4	23	0	2	0	0	2	25	14.49
1980	5	8	10	23	0	0	2	2	4	27	16.32
1981	6	4	2	12	0	2	0	1	3	15	9.32
1982	4	5	0	9	1	0	0	0	1	10	6.28
1983	5	4	5	14	0	3	0	1	4	18	11.39
1984	1	12	0	13	0	2	0	0	2	15	9.60
1985	2	0	0	2	0	0	0	0	2	2	1.33
Total	33	42	21	96	1	9	2	4	16	112	9.98

?? = Unknown sex

RESULTS

Secular Change of Incidence of Conjoined Twins

Table 1 shows the number of pre- and postnatal deaths from conjoined twins by sex of twin sets and the incidence rate during the period from 1979 to 1985; the overall proportion of postnatal deaths was 14% (16/112). The incidence of conjoined twins

was nearly constant except in 1985, whereas the highest rate (per million births) was in 1980 (16.32) and the lowest in 1985 (1.33). The incidence for this period was 9.98 per million births. The annual variations in the rate from 1979 to 1984 are not statistically significant ($\chi^2 = 9.82$, $df = 5$, $0.05 < P < 0.10$). The incidence for this period and 1985 was 11.3 and 1.33 per million births, respectively, the difference being significant ($P < 0.001$). Thirty-six of the 112 sets were males, 55 were females and 21 were unknown sexes. Excluding unknown sexes, female conjoined twins accounted for 60% of cases.

Table 2 - Numbers and incidence of conjoined twins according to maternal age, 1979-1985

Maternal age	Fetal deaths	Postnatal deaths	Total	Incidence (conjoined twins per million births)
Under 20	2	0	2	11.69
20-24	14	1	15	7.21
25-29	53	6	59	10.78
30-34	20	2	22	7.69
35-39	6	0	6	10.77
Over 40	1	1	2	25.04
Unknown	0	6	6	0.00
Total	96	16	112	9.89

Maternal Age and Birth Order

Table 2 shows the relationship between maternal age and the incidence of conjoined twins. The rate remained nearly constant except for age 40 years or more.

Table 3 - Conjoined twins and expected numbers according to birth order, 1979-1985

Birth order	Observed deaths from conjoined twins	Incidence (conjoined twins per million births)	Expected numbers based on uniform-birth-order incidence rate
1	42	9.27	45
2	34	7.95	42
3	22	13.46	16
4 +	8	24.77	3
Total	106	9.84	106

The rate per million births was lowest in mothers aged between 20 and 24 (7.2) and highest in mothers over the age of 40 (25.4).

Table 3 shows the relationship between birth order and the incidence on conjoined twins. Birth order was a numerical order in relation to all children liveborn or having died prenatally in the 21st week of gestation or later. The rate increased with birth order. The rate per million births was lowest in the second birth (8.0) and highest in birth order over 4 (24.8). The observed and expected number of cases with conjoined twins by birth order showed significant departure from homogeneity ($\chi^2 = 8.09$, $df = 2$, $P < 0.05$).

Table 4 - Conjoined twins according to month of conception and year

Year of birth	Month of conception												Total
	Jan.	Feb.	March	April	May	June	July	Aug.	Sept.	Oct.	Nov.	Dec.	
1979	3	3		1	1	2	5		5	3		2	25
1980	3			1	5	1	1	3	4	3	2		23
1981		1	1	1	1		1	2	3	3	1		14
1982	1	1		2	2			1			1	2	10
1983	2	1	2	2		1	2	4	1		1	1	17
1984	2	1	2	1	1	1	3			1	2	1	15
1985												2	2
Total	11	7	5	8	10	5	12	10	13	10	7	8	106

Seasonal Variation

The estimated month of conception for the 106 sets of twins is shown in Table 4. The month of conception was estimated in each case by counting backward in time from the data of birth, the calendar time equivalent of reported gestational length. Sixty (57%) twin sets were conceived during the months of July through December.

Geographical Variation

The figure shows the incidence of conjoined twins in each prefecture. The highest rate (per million births) was obtained in Gunma Prefecture (22.6), followed by Niigata (21.9) and Iwate (21.7), whereas the lowest rate was obtained in eleven prefectures (0), and Aichi (1.6). The regression coefficient for the incidence of conjoined twins on the latitude of the prefectural capital (per million degrees) was 0.615 ± 0.370 , which is not significantly different from zero. However, the incidence was slightly higher in the northeast than in the southwest.

The incidence of conjoined twins also computed by urban areas (ward and city) and rural areas (*gun*). The incidence was higher in urban areas (10.5 per million births) than in rural areas (8.2), but the difference was not significant.

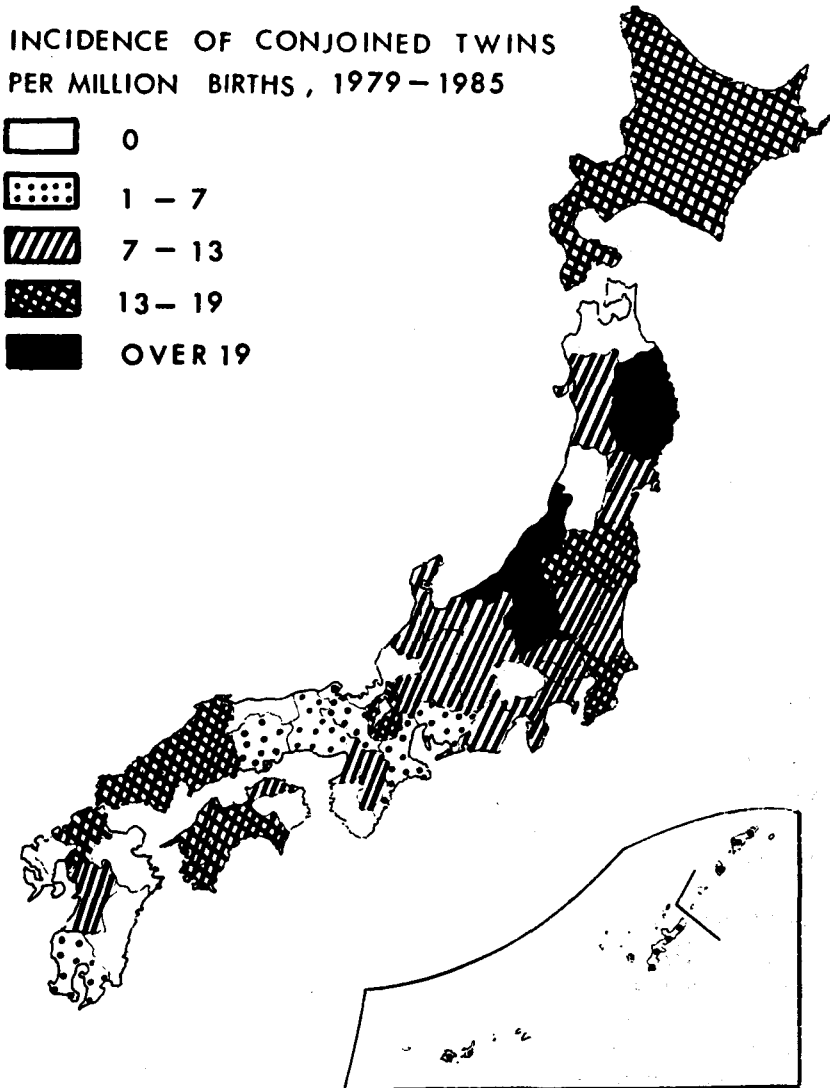


Figure. Incidence rates of conjoined twins, grouped into 5 broad categories, in each prefecture, 1979-1985.

Social Class Variation

The occupation of the head of household was classified into six groups by the classification on the deaths certificate: I (Agriculture only), II (Agriculture with other work), III (Self employed), IV (White collar), V (Blue collar) and VI (Other). Table 5 shows the number of twin sets and the incidence of conjoined twins by

occupation of the head of household. The incidence was highest in class I (14.4) and lowest in class VI (5.1). Table 5 also shows the expected number of deaths from conjoined twins by occupation of the head of household. The expected numbers were obtained by using a uniform social class incidence rate. The departure from homogeneity was not significant at the 5% level ($\chi^2 = 4.64$, $df = 4$).

Table 5 - Deaths from conjoined twins and incidence rate according to occupation of the head of household, 1979-1985

Occupation of head of household	Observed deaths from conjoined twins	Incidence (conjoined twins per million births)	Expected numbers based on uniform-birth-order incidence rate
I Agriculture only	4	14.38	3
II Agriculture with other works	9	9.93	9
III Self employed	13	10.14	12
IV Employee (white collar)	53	11.46	45
V Employee (blue collar)	28	8.19	34
VI Other	4	5.07	8
Total	111	9.89	111

DISCUSSION

Proportions of stillborn in conjoined twins were 40% (32 twin sets) in the United States [3], and 48% (15 twin sets) in South Africa [6], vs 85.6% in the present study. The former two studies used data from the Birth Defects Monitoring Program and the hospital record, respectively. In the present study, some conjoined twins could be still alive on the 31st of December in 1985. Accordingly, the rate of conjoined twins might be an underestimate of the true incidence in Japan. Milham [5] and Bender [1] reported the incidence of conjoined twins using data from the birth certificates, indicating the rates of 1:166,000 births and 1:200,000 live births, respectively. Edmonds and Layde [3] found 81 sets of conjoined twins in 7,903,000 births, giving a crude rate of 10.25 per million births. These authors suggest that a better estimate is between 1:100,000 and 1:30,000 births. In the present study, the rate of conjoined twins was 10.0 per million births.

According to Edmonds and Layde [3], no maternal-age effect was found in the United States. In the present study, a maternal-age effect was found in mothers over the age of 40, where the highest rate was obtained.

There was no seasonal variation in the time of conception in Southern Africa [6] and the United States [1]. On the other hand, Milham [5] reported that 77% of twins (17 twin sets) were conceived during the months of July through December. In the present study, the corresponding rate was 57%. However, these data on conjoined twins were quite small for determining seasonal variations.

REFERENCES

1. Bender C (1967): Studies on symmetrically conjoined twins. *J Pediatr* 70:1010-1011.
2. Bhattay E, Beighton P, Nelson MM (1975): Epidemic of conjoined twins in Southern Africa? *Lancet* ii:741-743.
3. Edmonds LD, Layde PM (1982): Conjoined twins in the United States, 1970-1977. *Teratol* 25:301-308.
4. Japan, Ministry of Health and Welfare (1981-1987): Vital statistics, 1979-1985. Health and welfare Statistics and Information Department, Ministry of Health and Welfare, Tokyo (in Japanese).
5. Milham S (1966): Symmetrical conjoined twins: An analysis of the birth records of twenty-two sets. *J Pediatr* 69:643-647.
6. Viljoen DL, Nelson MM, Beighton P (1983): The epidemiology of conjoined twinning in Southern Africa. *Clin Genet* 24:15-21.
7. Zake EZN (1984): Case reports of 16 sets of conjoined twins from a Uganda hospital. *Acta Genet Med Gemellol* 33:75-80.

Correspondence: Dr. Y. Imaizumi, Institute of Population Problems, 2-2 1-Chome, Kasumigaseki, Chiyoda-Ku, Tokyo, Japan.