The influence of the birth of a malformed child on the mother's further reproduction

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Record, R. G. and Armstrong, E. (1975). *British Journal of Preventive and Social Medicine*, 29, 267-273. **The influence of the birth of a malformed child on the mother's further reproduction.** Births which occurred in Birmingham in 1964-70 were assembled into fraternities by computer linkage. By calculating the frequency with which one birth was followed by another and the interval between births the reproductive behaviour of mothers after the birth of a malformed child was compared with that of all mothers, taking account of differences in maternal age, parity, and period of observation. It was found that malformations which resulted in stillbirth or early death were more frequently followed by another birth and that the interval to the following birth was shorter than usual. In this respect malformations did not differ in their effect from other causes of stillbirth and infant death. The birth of children with severe malformations who survived, however, acted as a slight deterrent to further reproduction. The malformation rate among children born after a malformation was double the usual rate; the recurrence rate was particularly high for neural tube defects, 5% of subsequent children being affected. In spite of this parents of children with central nervous system malformations were not deterred from further reproduction unless the affected child survived.

There have been only a few studies of the attitude of parents with regard to further reproduction after the birth of a malformed child. Specific defects which have been investigated from this point of view include spina bifida (Hare, *et al.*, 1966; Timson, 1970; Walker, Thomas, and Russell, 1971; Richards and McIntosh, 1973; Hunt, 1973), congenital heart disease (Polani and Campbell, 1955; Apley, Barbour, and Westmacott, 1967; Boon, 1972), and Down's disease (Newcombe, 1965). There are also a few references to reproductive behaviour following stillbirths and infant deaths (Newcombe and Rhynas, 1962; Sheps, 1965; Vogel and Knox, 1975) where no attempt was made to distinguish between malformations and other causes of death.

The impression gained by reading these accounts is that the birth of a malformed child may affect the subsequent reproductive behaviour of parents in various ways. Some parents may be so shocked by the event that they may resolve to have no more children in order to avoid any risk of enduring again such a distressing experience. Indeed this

course may be urged upon them by genetic counsellors and other advisors in the case of malformations such as neural tube defects where the risk of recurrence may be regarded as unacceptably high. On the other hand some parents may respond by deciding to have another child as quickly as possible to compensate for the disappointment caused by the birth of a defective child. This may be more likely if the malformed child died at or soon after birth. If however the child lives but is severely disabled requiring constant maternal attention the parents may postpone further reproduction for an indefinite period, possibly for ever.

Some of these views are based on parents' stated intentions shortly after the malformed birth and others result from following-up parents for varying periods and noting the number of subsequent children or, in some cases, the total size of the completed family. Interpretation of some of these studies is hampered by the absence of an acceptable control group. There seems to be no record of a comprehensive study of malformed births designed

to investigate whether further reproduction is influenced by the type of malformation and its outcome in terms of early death or severe handicap. We were therefore prompted to examine the possibility of using the Birmingham malformation register and the associated birth data for this purpose. The present paper describes the method and results of this investigation.

METHOD

Data relating to all births to Birmingham residents have been collected continuously since 1950. In 1964 a reliable means of maternal identification was introduced based on the mother's date of birth, the first two letters of her forename, and the first three letters of her maiden surname. By means of a simple computer linkage operation it was possible to assemble in sequence, data on all births to each woman from the beginning of 1964 to the end of 1970. The method worked well with the records of the indigenous population but immigrants presented some difficulty because some did not know their exact date of birth and some had surnames which were not sufficiently distinctive. It was decided therefore to restrict the analysis to mothers of British, Irish, or European origin. The effect on later reproduction of twins in a fraternity is itself of interest and will be reported on later. Their inclusion here, however, complicates the analysis and it was thought that little error would result from excluding them. The present analysis is, therefore, based on Birmingham women of Caucasian origin who had singleton births in the years 1964-69 followed-up to the end of 1970. The series consists of 69 196 mothers and 97 467 births, but for 317 births the age or parity of the mother was not known. These were excluded leaving 97 150 births of whom 2169 were malformed (22 per 1000).

The successful assembling of fraternities depends on the continued residence within the city of each mother and since about 35% of children born in 1964 were not traceable at the age of five years it is clear that a number of families had left the city during the follow-up period and some of their subsequent births would not have been recorded. The present study, therefore, understates the reproductive experience of the women in the series. Nevertheless comparison of a special group of women, for example those who had a malformed child, with the whole population of mothers is feasible provided they do not differ from other mothers in their mobility. The methods of measuring reproductive behaviour which we adopted enabled us to investigate this point.

There are several ways of measuring reproductive performance of a group of mothers subsequent to a particular birth. The two simplest measures, and the ones chosen for this study are:

- (1) the proportion of women who have a child subsequent to the index birth within a stated period (in this case up to the end of 1970);
- (2) for those who have a subsequent birth, the interval from the index birth to the next birth following.

There is particular advantage in using both these measures in a series subject to loss by movement out of the study area. It is possible that families in which a stillbirth or early death occurs are more mobile than those where the child survives, and if a higher proportion than average of these families move out of the area, so that subsequent births do not come to notice, the proportion of mothers for whom a later birth is recorded would be less than for less mobile families. This would give an impression of impaired reproductive performance and such a group of women would be expected to show a longer than average interval before the next birth. In fact, because women moving out of the area have a shorter time in which to have their next baby if it is to be included in the study the average interval would tend to be less than normal.

If, therefore, a group of women showed a paradoxical result of fewer subsequent births than usual and a shorter interval between births, one would suspect undue movement of women out of the series and no conclusion concerning their reproductive performance would be permissible. On the other hand, consistency between the two measures—(a) a smaller proportion of mothers having subsequent births and a longer interval before the next birth, or (b) more mothers having later births and a shorter interval—would suggest respectively impaired or enhanced reproductive performance.

In using these measures of reproductive behaviour to compare a particular group of mothers with all mothers, it is necessary to take account of three important variables which are closely related to reproductive performance—age of the mother, the number of previous children, and the duration of follow-up (which depends of course on the year of the index birth). The effect of these variables on the proportion of index births followed by a later birth is shown in Table I. As would be expected, the proportion falls with increasing age and with reduction in the duration of follow-up, but the relation to the number of previous births is more complicated. When the simultaneous relationship to all three variables is studied a very complex

TABLE I
PROPORTION OF BIRTHS FOLLOWED BY A LATER BIRTH AND MEAN INTERVAL FROM INDEX BIRTH TO NEXT FOLLOWING

Births	Age of Mother (Years)					Total
	Under 20	20-24	25-29	30-34	35 and over	
Number	11202	33654	26332	15037	10925	97150
Number followed by a later birth	4545	11920	7244	3161	1364	28234
Percentage followed by a later birth	40.6	35.4	27.5	21.0	12.5	29.1
Mean interval (days)	743	801	838	842	800	806

Births	Number of Previous Births					Total
	0	1	2	3	4 and over	
Number	35110	27626	15518	8287	10609	97150
Number followed by a later birth	13256	6839	3558	1928	2653	28234
Percentage followed by a later birth	37.8	24.8	22.9	23.3	25.0	29.1
Mean interval (days)	802	817	808	810	792	806

Births	Year of Birth						Total
	1964	1965	1966	1967	1968	1969	
Number	17866	17198	16456	16124	15399	14107	97150
Number followed by a later birth	7125	6506	5694	4546	3177	1186	28234
Percentage followed by a later birth	39.9	37.8	34.6	28.2	20.6	8.4	29.1
Mean interval (days)	936	883	812	715	606	455	806

317 births for which the age of the mother or the number of previous births was not known have been excluded

pattern is revealed, the proportion ranging from 58% for mothers under 20 in 1964 with no previous births to less than 2% for mothers of 35 and over in 1969 with two previous births. Table I also shows that the interval between births is related to age and parity, and year of birth. The interval increases with increasing age up to 35 and then falls, and decreases steadily as the duration of follow-up decreases. The relation to parity is less regular.

It follows that simultaneous standardization for the three variables is a necessary preliminary to any comparison between mothers of malformed children and other mothers. This can be done if the total population of births is allocated according to maternal age (five groups), parity (five groups), and year of birth (six years). This grouping produces 150 cells. The proportion of mothers who had a subsequent birth and the mean interval from the index birth to the next following are calculated for each cell. Expected values for any specified malformation group can then be derived by summation over the whole table taking account of the number of malformed births in each age/parity/year of birth cell. Comparison of the observed and expected

values gives a measure of the relative reproductive performance of mothers after the malformed birth.

RESULTS

Since some malformations frequently result in death at or soon after birth it seemed desirable, before examining the effect of the birth of a malformed child on subsequent reproduction, to investigate whether a perinatal death or a death later in infancy influenced the parents' reproductive pattern. Results of this analysis indicate that both perinatal and later infant deaths are followed by an increased likelihood of a subsequent birth and that the interval before the next birth is shorter than usual (Table II). This suggests that parents who have been disappointed in the outcome of a pregnancy by stillbirth or death of the child tend to compensate for the loss. In view of this it is necessary in the analysis which follows to distinguish between malformations which resulted in stillbirth or death in infancy and those where the child survived the first year. It also seems advisable to distinguish between major malformations which threaten survival or seriously impair the quality of life and

TABLE II

REPRODUCTIVE EXPERIENCE OF WOMEN FOLLOWING A PERINATAL DEATH (STILLBIRTH OR DEATH IN THE FIRST WEEK) AND A DEATH IN THE REST OF THE FIRST YEAR

Deaths	No.	No. followed by a Subsequent Birth			Mean Interval to Next Birth (Days)		
		Observed (O)	Expected* (E)	O/E	Observed (O)	Expected* (E)	O/E
Perinatal	2053	745 (36.3)	589 (28.7)	1.26	681	787	0.87
Infant after first week . .	698	293 (42.0)	211 (30.2)	1.39	691	801	0.86
Total	2751	1038 (37.7)	800 (29.1)	1.30	684	791	0.86

*Expected values are based on the experience of all mothers distributed by maternal age, parity, and year of index birth as the mothers of dead infants
Percentages are given in parentheses

TABLE III

REPRODUCTIVE EXPERIENCE OF WOMEN FOLLOWING THE BIRTH OF CHILDREN WITH MAJOR MALFORMATIONS RESULTING IN STILLBIRTH OR INFANT DEATH

Site and Type of Malformation	No. of Cases	No. Followed by Another Birth			Mean Interval to Next Birth (Days)		
		Observed (O)	Expected (E)	O/E	Observed (O)	Expected (E)	O/E
<i>Nervous system</i>							
Anencephalus	134	55	37.15	1.48	711	787	0.90
Spina bifida, encephalocele, iniencephalus	102	35	29.12	1.20	680	797	0.85
Hydrocephalus	35	15	9.97	1.50	476	772	0.62
<i>Heart and great vessels</i> . .	174	75	50.54	1.48	693	801	0.87
<i>Upper alimentary tract</i>							
Clefts of lip and palate . .	14	6	4.73	1.13	1000	887	0.92
Tracheo-oesophageal fistula	11	3	3.27		476	931	
<i>Lower alimentary tract</i>							
Anal atresia	13	5	3.54	1.24	428	822	0.72
Other intestinal obstruction (excluding meconium ileus)	7	2	2.12		737	463	
<i>Genito-urinary tract</i>							
Renal agenesis	18	7	4.04	1.55	512	664	0.82
Other major defects	16	7	4.98		657	755	
<i>Other abdominal</i>							
Diaphragmatic hernia . . .	17	7	4.93	1.07	553	734	0.73
Hiatus hernia	1	0	0.31				
Exomphalos	13	2	3.16		455	722	
<i>Down's disease</i>	40	12	8.57	1.40	692	841	0.82
<i>Other</i>	40	14	12.32	1.14	593	746	0.79
Total	635	245	178.75	1.37	664	788	0.84

minor malformations which are easily treated or require no treatment and which result in little or no disability.

There were 635 infants with major malformations who were born dead or died before their first birthday (Table III). Of these, 245 were followed by another birth within the period of the study. The number 'expected', based on the experience of all mothers of similar age and parity was 179 and the ratio of observed to expected was 1.37. The mean interval between the birth of the malformed child and the next birth was 664 days. The expected mean interval was 788 days giving an observed to expected ratio of 0.84. These ratios closely resemble those shown in Table II based on stillbirths and

infant deaths from all causes. This suggests that if the child died, the fact that it was malformed did not deter parents from further reproduction and they tended to hasten the next pregnancy as if the dead child had not been malformed.

Table III also shows the malformations divided according to site and type. Infants with two or more malformations have been assigned to the more serious category. For example, cases of anencephalus with spina bifida have been allocated to anencephalus, and renal agenesis with diaphragmatic hernia to renal agenesis. All cases of Down's disease have been included under that heading even if cardiac defects or other malformations were present.

When divided in this way some categories contain

very small numbers. Nevertheless they showed considerable consistency; for every group there were more subsequent births than expected and the mean interval from the malformed birth to the next was shorter than usual. In the case of malformations of the nervous system and of the heart numbers are large enough to establish these points with some confidence.

Infants with major malformations who survived the first year numbered 656 of whom 165 had been followed by another birth by the end of the study. The number expected was 180 giving an observed to expected ratio of 0.92. (Table IV). The mean interval from the malformed birth to the next was 863 days. The expected mean interval was 829 days and the ratio of observed to expected was 1.04. This result suggests that the continued presence in the home of an infant with a major malformation is a slight deterrent to further reproduction by the parents but the effect is small and cannot be regarded as established beyond doubt in this series. The individual groups of malformations can be viewed with even less confidence but there is a suggestion that spina bifida has a slightly greater deterrent effect than the other types.

There were 848 infants with minor malformations who survived the first year. The number followed by another birth (244) was very close to that expected (248) and the mean interval between the index birth

and the next was not very different from that expected (Table V). It appears that minor malformations which carry no risk of death or serious disability do not lead to any modification of the parents' reproductive behaviour.

There were 30 infants with minor malformations who died from causes unconnected with the malformation. Eleven were followed by another birth (expected 10.1) and the mean interval was 930 days (expected 708 days). In view of the small numbers little importance can be attached to this result.

DISCUSSION

The results of this investigation suggest that the birth of a severely malformed child acts as a slight deterrent to further reproduction if the child survives. This is perhaps hardly surprising in view of the exacting nature of the task of caring for children with these defects and the anxiety of parents regarding the outcome. On the other hand if the child is stillborn or dies early in life the parents tend to compensate for the loss in the same way that they do when a non-malformed child dies. This result was somewhat unexpected since it was thought that parents of children with certain types of malformation, particularly neural tube defects would have been told of the increased risk to subsequent children. In view of this it seemed worth

TABLE IV

REPRODUCTIVE EXPERIENCE OF WOMEN FOLLOWING THE BIRTH OF CHILDREN WITH MAJOR MALFORMATIONS WHO SURVIVED THE FIRST YEAR

Site and Type of Malformation	No. of Cases	No. Followed by Another Birth			Mean Interval to Next Birth (Days)		
		Observed (O)	Expected (E)	O/E	Observed (O)	Expected (E)	O/E
Nervous system							
Spina bifida, encephalocele, iniencephalus ..	107	26	31.71	0.82	866	814	1.06
Hydrocephalus ..	30	11	9.57	1.15	859	854	1.01
Heart and great vessels ..	125	31	34.88	0.89	787	833	0.94
Upper alimentary tract							
Clefts of lip and palate ..	133	34	35.42	0.96	943	844	1.12
Tracheo-oesophageal fistula	12	3	3.07		971	840	
Lower alimentary tract							
Anal atresia ..	22	6	5.91	1.18	641	811	1.08
Other intestinal obstruction	8	3	1.17		1350	806	
Genito-urinary tract ..	24	6	6.38	0.94	798	743	1.07
Other abdominal							
Diaphragmatic hernia ..	1	0	0.05	0.80	—	—	1.41
Hiatus hernia ..	9	2	2.46		1340	902	
Exomphalos ..	5	1	1.22		898	741	
Down's disease ..	109	21	23.53	0.89	791	827	0.96
Limbs							
Reduction deformities ..	30	10	10.15	0.99	880	874	1.01
Other ..	41	11	13.83	0.80	844	807	1.05
Total ..	656	165	179.89	0.92	863	829	1.04

TABLE V
REPRODUCTIVE EXPERIENCE OF WOMEN FOLLOWING THE BIRTH OF CHILDREN WITH MINOR MALFORMATIONS WHO SURVIVED THE FIRST YEAR

Site and Type of Malformation	No. of Cases	No. Followed by Another Birth			Mean Interval to Next Birth		
		Observed (O)	Expected (E)	O/E	Observed (O)	Expected (E)	O/E
Special senses							
Eye	10	2	3.40	0.77	680	888	1.05
Accessory auricle	40	8	11.47		963	760	
Other ear	18	5	4.55		742	855	
Genito-urinary tract							
Hypospadias	75	25	20.84	1.09	802	785	1.02
Other	10	0	2.19		—	—	
Limbs							
Club foot	327	81	91.81	0.99	680	782	0.95
Polydactyly	86	24	25.98		832	802	
Syndactyly	79	24	23.81		862	825	
Congenital dislocation of hip	153	59	48.07		828	848	
Other	13	5	4.97		649	727	
Other	37	11	10.45	1.05	688	807	0.85
Total	848	244	247.54	0.99	771	806	0.96

while to examine the data further to ascertain the malformation rate among children born after the index case.

It was shown in Tables III and IV that the series contained 1291 major malformations of which 410 were followed by a later birth. Eighteen of these later births had severe malformations and four had minor defects—a total incidence of 54 per 1000 which is more than double the rate for the series as a whole. Details are shown in Appendix I. There is a high recurrence rate for neural tube defects which were present in seven of the 142 infants born after a malformation of the central nervous system. This corresponds to a rate of 49 per 1000 which is more than ten times the rate for the whole series (4.2 per 1000). This is in accord with previous estimates of the recurrence rate of neural tube defects (Carter, Laurence, and David, 1968). It is not clear if parents received little information about the possibility of these malformations recurring in later pregnancies or whether they considered the risk to be within acceptable limits.

Although the other malformation groups showed less tendency to specific recurrence the malformation risk in subsequent pregnancies is certainly greater than normal. In particular there seems to be some evidence of an association within sibships between congenital heart disease and clefts of lip and palate, both types of defect being noted in each of four fraternities.

The malformation rate among infants following births with minor defects was even higher than the rate after major malformations, 18 of the 244 children being affected (74 per 1000), but in only

four was the second malformation regarded as severe. In ten instances the type of defect was the same as in the preceding birth (Appendix II). Although the risk of recurrence cannot be disregarded most of these minor malformations are amenable to treatment and produce little or no disability. It seems reasonable therefore that parents who have had an affected child should not be advised against having more children and it is clear from the results shown in Table V that parents were not deterred by the occurrence of these malformations.

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APPENDIX I

FREQUENCY OF MALFORMATIONS AMONG INFANTS FOLLOWING THE BIRTH OF CHILDREN WITH MAJOR MALFORMATIONS

Site and Type of Malformations (Index Birth)	No. Followed by Another Birth	No. Malformed	Type of Malformation
Nervous system			
Anencephalus	55	3	Anencephalus (1) Spina bifida (2)
Spina bifida, encephalocele	61	3	Anencephalus (2) Aortic anomaly (1)
Hydrocephalus	26	2	Hydrocephalus (1) Microcephalus (1)
Heart and great vessels	106	7	Valve defect (1) Cleft lip (1)
			Cleft palate (1) Hermaphrodite (1)
			Hypospadias (1) Club foot (1)
			Polydactyly (1)
Alimentary tract			
Clefts of lip and palate	40	2	Congenital heart defect (2)
Other	22	0	
Genito-urinary tract	20	1	Diaphragmatic hernia and renal hypoplasia (1)
Down's disease	33	1	Craniorachischisis (1)
Other	47	3	Transposition of great vessels (1)
			Laurence-Moon-Biedl syndrome (1)
			Club foot (1)
Total	410	22	

APPENDIX II

FREQUENCY OF MALFORMATIONS AMONG INFANTS FOLLOWING THE BIRTH OF CHILDREN WITH MINOR MALFORMATIONS

Type of Malformation (Index Birth)	No. Followed by Another Birth	No. Malformed	Type of Malformation
Defects of external ear	13	4	Polycystic kidney (1) Microtia and accessory auricle (1)
			Hypoplasia of pinna (1) Club foot (1)
Hypospadias	25	2	Cleft lip and palate (1) Hypospadias (1)
Club foot	81	6	Down's disease (1) Hypospadias (1) Club foot (4)
Polydactyly	24	1	Talipes (1)
Congenital dislocation of hip	59	4	Cri-du-chat syndrome (1) Congenital dislocation of hip (3)
Other limb defects	29	1	Megacolon and polydactyly (1)
Other defects	13	0	
Total	244	18	