

The epidemiology of sudden infant death syndrome

J. F. MURPHY

From the Department of Child Health, University Hospital of Wales, Cardiff

R. G. NEWCOMBE

From the Department of Medical Statistics, Welsh National School of Medicine, Cardiff

J. R. SIBERT

From the Llandough Hospital, Penarth, South Glamorgan

SUMMARY Ninety-nine cases of sudden infant death syndrome were identified among 47 413 liveborn deliveries to Cardiff residents during the years 1965-73 and 1975-77. Nineteen predictive factors available on the Cardiff Birth Survey record were evaluated individually and jointly in terms of their power to identify risk of sudden and unexpected infant death.

In a large proportion of postneonatal infant deaths, clinical history and post-mortem examination do not reveal pathological conditions sufficient to account adequately for the death. In a prospective study carried out by the working party for early childhood deaths in Newcastle,¹ three-quarters of the postneonatal deaths studied were classified as sudden and unexpected. The loss of an infant in this way is particularly hard for the parents to bear, because in the absence of a clearcut physiological explanation they commonly feel the cause must lie in what they have or have not done; such feelings of guilt may be shared by members of the attending primary care team.² The fact that most postneonatal infant deaths defy adequate explanation constitutes a major defect in the body of medical knowledge.

A major problem hindering research arises from the essentially negative nature of the diagnosis: observer differences in completeness of necropsy technique and interpretation and in certification of the cause of death.³⁻⁴ Two main areas of research are related to sleep apnoea and associated cardio-respiratory pathophysiology in young infants⁵⁻⁷ and the study of epidemiological data.^{8,9} We have examined the medical and social factors of all Cardiff infants and established a scoring system weighting these factors in such a way as to discriminate infant deaths attributed to sudden infant death syndrome (SIDS) from all other live births.

Material and methods

The Cardiff Birth Survey records demographic, social, and medical information on all births to Cardiff residents.¹⁰ We examined all such births occurring during the 12 years 1965-1973 and 1975-1977; the year 1974 was excluded because of incomplete ascertainment of cases.

Every infant under the age of 1 dying suddenly and unexpectedly at home, who had been well before death, underwent post-mortem examination by the duty consultant pathologist. The post-mortem technique did not include any additional specialised biochemical, virological, or histological procedures. Among such infants, those without congenital abnormalities or any other pathological findings sufficient to account for death were included in the SIDS group. In the early years of the study non-specific diagnoses such as 'mild bronchiolitis' had been used; latterly the expression SIDS was used by all pathologists.

The control group consisted of all remaining liveborn infants born to Cardiff residents during the study period.

The incidence of SIDS was tabulated by all variables recorded by the Cardiff Birth Survey. Nineteen variables were identified which gave independent information bearing on prediction. Certain factors were excluded in favour of other

highly correlated but more discriminatory variables: duration of marriage, history of abortion, inadequate assessment and correction of maternal anaemia, gestational age at delivery, neonatal infection and recourse to antibiotic therapy. Birth weight was a relatively weak predictor on the present data and a case could be made for its exclusion also; nevertheless it was included in the predictor because of its importance in other studies and its widespread acceptance as an easily determined indicator of prognosis. Numerous other variables examined did not significantly correlate with SIDS: maternal height and weight, marital status, medical, obstetric and oral contraception history, type of antenatal care, use of ultrasound and cervical cerclage, excess weight gain during pregnancy, toxæmia, bacilluria, version, presentation, Rhesus sensitisation, place of delivery, presence of a doctor at delivery, use of oxytocin or prostaglandin, anaesthesia or analgesia, scalp sampling, and the Apgar score.

Table 1 lists the 19 predictive variables thus selected. The 19 wards of the City of Cardiff were segregated into three groups according to the type of housing in an arbitrary way before the data were examined. Three wards could be unequivocally labelled 'good', consisting almost entirely of well-maintained owner-occupied property. The wards termed 'poor' consisted almost entirely of either old or council housing. The intermediate group included areas of an intermediate standard and others whose boundaries contained widely different standards of housing. 'Complications of pregnancy' means the occurrence of any of the following: antepartum haemorrhage; rupture of membranes 24 hours before the onset of labour; venereal disease; deep-vein thrombosis or embolism. Season of birth was dichotomised in January and July after examining the data and the true significance of the effect was confirmed using a more appropriate Edwards test.¹¹

In Table 1 the relative risk (RR) is obtained by dividing the incidence of SIDS in a subgroup by that in the whole series. This definition of RR, which differs from that commonly used in epidemiology, is appropriate in the present context (Newcombe RG, unpublished observations). Each RR is then converted to an additive score component of $10 \log_{10} RR$, rounded to the nearest whole number.

In this way a scoring system, system I, was set up, which could allocate to any infant a score representing the risk of SIDS. An infant is given an initial score of 100, and 19 components, one for each factor, are added to or subtracted from this baseline. A zero score is given to a factor whose value is not known.

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The distribution of each score component and the total score among the 99 cases and 47 314 controls were examined. Assessment of discriminatory power was made in terms of a statistic P, the probability that a randomly selected case of SIDS would have scored higher than a control selected independently at random.¹²⁻¹⁴ The off-diagonal entries of the correlation matrix of the 19 score components among all 47 413 infants were examined, in order to confirm that assumptions of independence were not substantially violated.

A second scoring system was constructed, using equal weights. In this analysis each score of +1 or above was replaced by +1 and each zero or negative score was replaced by zero.

A third scoring system was also derived, based on a minimal set of salient variables: maternal smoking, maternal age, area of residence, and birth weight, each given a weight as in system I.

Results

Ninety-nine cases of SIDS were identified among 47 413 liveborn infants, an incidence of 2.1 per 1000 live births. The incidence is cross-tabulated with the 19 predictive factors in Table 1. These are arranged in decreasing order of predictive power; values of P for smoking and employment of partner are 0.647 and 0.520.

The 19 score components are not strongly correlated with one another: 137 of the 171 entries in the correlation matrix are less than 0.1 in absolute value.

Table 2 indicates the mean, SD, and range of the total score, and the value of P, for systems I, II, and III. It can be seen that some information is lost in simplifying system I to either II or III. System I assigns a score of 107 or higher to 5057 or 11% of the 47 413 infants, and these include 49% of the cot deaths (see Figure).

Discussion

This study demonstrates that an infant from a certain type of family has a higher than average risk of suffering a cot death—essentially a deprived family of low social class living in a poor area. The mother is typically young, and yet of relatively high parity; many mothers in our study smoked and were poor clinic attenders. Labour was easy, with a rapid second stage; the infant was more likely to be male or twin and to have spent some time in the special care baby unit. Although none of these factors adequately explains the death, their appearance as epidemiological factors does seem plausible.

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Table 1 *Factors differing significantly between 99 SIDS and 47 314 control births*

<i>Factor</i>	<i>Cases</i>	<i>Controls</i>	<i>Incidence per 1000</i>	<i>RR</i>	<i>Score</i>	<i>Significance level</i>
SMOKING HABITS OF MOTHER						
None/ex-smoker	32	27 031	1.18	0.57	-2	0.001
Under 20 a day	39	14 970	2.60	1.24	+1	
20 or more a day	25	4 421	5.62	2.69	+4	
AGE OF MOTHER AT BIRTH						
Under 18	12	1 881	6.33	3.04	+5	0.001
18 to 24	62	21 302	2.90	1.39	+1	
25 or over	24	23 927	1.04	0.50	-3	
AREA OF RESIDENCE						
Good	1	4 060	0.25	0.12	-9	0.001
Medium	35	25 357	1.38	0.66	-2	
Poor	63	17 897	3.51	1.68	+2	
SOCIAL CLASS						
I or II	2	7 070	0.28	0.14	-9	0.001
III or IV	64	30 654	2.08	1.00	0	
V	18	4 741	3.78	1.81	+3	
ANTENATAL CLINIC ATTENDANCES						
Under 3	11	1 726	6.34	3.04	+5	0.001
3 or 4	13	3 837	3.38	1.62	+2	
5 to 14	68	34 031	1.99	0.96	0	
15 or more	5	6 835	0.73	0.35	-5	
2nd STAGE OF LABOUR (MINS)						
Under 15	47	16 660	2.81	1.35	+1	0.025
15 to 19	30	13 049	2.29	1.10	0	
30 to 59	13	9 854	1.32	0.63	-2	
60+	3	4 235	0.71	0.34	-5	
EMPLOYMENT OF MOTHER						
None	81	30 645	2.63	1.26	+1	0.05
Non-sedentary ending before 20 wks	} 11	8 233	1.33	0.64	-2	
Sedentary ending before 20 wks						
Non sedentary continuing after 20 wks	} 5	7 262	0.69	0.33	-5	
Sedentary continuing after 20 wks						
COMPLICATIONS OF PREGNANCY						
Present	32	7 169	4.44	2.13	+3	0.001
Absent	66	39 311	1.68	0.80	-1	
MATERNAL BIRTH INJURY						
None	58	20 665	2.80	1.34	+1	0.01
Episiotomy	31	18 075	1.71	0.82	-1	
More serious injury	10	8 320	1.20	0.57	-2	
INFANT FEEDING						
Wholly or partly breast-fed	16	14 223	1.12	0.54	-3	0.005
Lactation suppressed or not tried	72	27 574	2.60	1.25	+1	
PREVIOUS DELIVERIES						
None	26	17 655	1.47	0.70	-2	0.05
One	31	14 461	2.14	1.02	0	
Two or more	42	14 998	2.79	1.34	+1	
SEASON OF BIRTH						
August to January	62	23 002	2.69	1.29	+1	0.01
February to July	37	24 312	1.52	0.73	-1	
GESTATION AT BOOKING						
Under 20 weeks	67	28 740	1.73	0.83	-1	0.001
20 weeks or over	30	8 043	3.72	1.78	+3	
SEX OF INFANT						
Male	63	24 114	2.61	1.25	+1	0.02
Female	36	23 199	1.55	0.74	-1	
ADMISSION TO SCBU						
No	79	43 392	1.82	0.87	-1	0.001
Yes	20	3 922	5.07	2.43	+4	
MODE OF DELIVERY						
Spontaneous	90	37 845	2.37	1.14	+1	0.01
Forceps, breech or caesarean section	9	9 469	0.95	0.45	-3	
BIRTHWEIGHT						
Under 2500g	15	3 108	4.80	2.30	+4	0.005
2500g or more	83	44 164	1.88	0.90	0	
MULTIPLICITY						
Singleton	90	46 371	1.94	0.93	0	0.001
Multiple	9	943	9.45	4.53	+7	
EMPLOYMENT OF PARTNER						
Employed	89	44 912	1.98	0.95	0	0.05
Unemployed	6	1 090	5.47	2.62	+4	

Table 2 Performance of scoring systems to predict sudden infant death syndrome

System	No. of factors	Weights	Group	Mean	SD	Range	P
I	19	Unequal	Cases	106.4	9.0	85-126	0.816
			Controls	93.6	10.9	48-133	
II	19	Equal	Cases	108.8	2.3	103-114	0.786
			Controls	106.1	2.3	100-116	
III	4	Unequal	Cases	102.3	4.5	86-111	0.750
			Controls	97.9	4.9	86-115	

Infants born between August and January are at highest risk because they reach the most susceptible age during the winter months. The effect of maternal smoking may be understood in terms of either damage in utero or passive smoking by the neonate.

The measure of predictive power used, P, takes the value 1.0 when discrimination is perfect, 0.5 when the distributions in cases and controls are indistinguishable. The level of prediction obtained, 0.816, is similar to that found by Carpenter *et al.*⁹

We compared the scoring system with that obtained by studying a group of 80 Cardiff children who had suffered non-accidental injury (NAI). Murphy *et al.*¹⁵ reported such a system, based on the Cardiff Birth Survey, using equal weights. For purposes of cross-validation an unequally weighted version of the NAI score was used. The SIDS and NAI scores had eight factors in common, which were predictive of both outcomes: maternal age, social class, city ward, maternal smoking, poor use of antenatal care, lack of employment during pregnancy, admission to the special care baby unit, and artificial feeding. Birth weight was predictive of NAI and was included in the SIDS predictor. Two further factors related to NAI were marital status and low maternal weight. The NAI predictor can predict NAI with a power of $P = 0.858$ and SIDS with a power of $P = 0.768$. Conversely the SIDS score predicts SIDS and NAI with $P = 0.816$ and 0.756 respectively. Thus the two predictors do not delineate quite the same high-risk group but they are in close agreement. On epidemiological and social grounds much the same group of infants is at high risk of both SIDS and NAI.

These findings support those of Roberts *et al.*¹⁶ who demonstrated a threefold increased risk of infant death among siblings of abused children. In our study a high-risk group consisting of 11% of all infants who score 107 or more would include 49% of the cases of SIDS. Half the fatalities would occur in the low-risk group and only 1% of the high-risk group would be expected to die. This relatively weak predictive

power renders any formal follow-up system for high-risk infants very costly in manpower and would be expected to make little impact on the incidence of SIDS. Such a prediction system is, however, a valuable means of identifying a group of infants on which to base further research.⁵ It may also be used in health care planning to delineate those city wards and families which may benefit most from additional supportive health services.¹⁷

The predictive power is unlikely to improve until it is possible to identify risk factors of a physiological rather than a social nature. This study does, however, once again underline the impact of the social circumstances of the family on the well-being of the infant.

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Reprints from Dr. R. G. Newcombe, Department of Medical Statistics, Welsh National School of Medicine, Heath Park, Cardiff CF4 4XN.

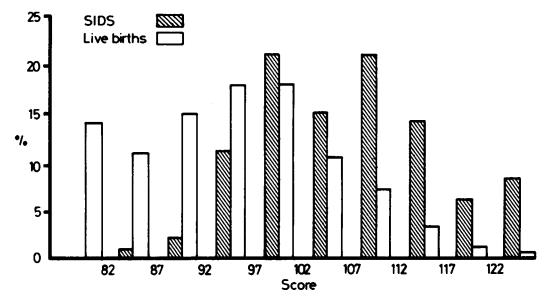


Figure Distribution of score 1 in cases of sudden infant death syndrome and in all live births.

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