Sudden infant death in Southampton and an evaluation of the Sheffield scoring system

J D C HARRIS,¹ M RADFORD,² M WAILOO,² R G CARPENTER,³ AND D MACHIN⁴

From the Faculty of Medicine,¹ Child Health,² and Community Medicine,⁴ University of Southampton, Southampton, and Department of Epidemiology and Medical Statistics,³ London School of Hygiene and Tropical Medicine, London, UK

SUMMARY The cases of sudden infant death occurring in Southampton over the period 1 July 1975 to 31 December 1979 are reviewed with respect to “risk” factors identified in other studies. A retrospective case control study of 49 of these deaths and 98 controls was undertaken to evaluate the Sheffield scoring system. The study suggested that the Sheffield score might usefully be used prospectively in Southampton with only minor modification.

The development of a method for predicting at birth which babies are liable to die suddenly and unexpectedly at home¹ has raised hopes of reducing the postneonatal infant mortality rate. By giving numerical weightings to eight obstetric and perinatal factors a score for each baby is obtained² that is related to the risk of unexpected death and in conjunction with health visitor surveillance has resulted in a reduced number of unexpected infant deaths in Sheffield (RG Carpenter *et al*, unpublished observations).

The value of score systems in general has been questioned³ as has the applicability of the Sheffield system to other areas.^{4,5} Recent work has shown, however, that although the Sheffield system may not be directly applicable in other areas, minor modifications may yield a useful system.⁶

The purpose of this paper is to describe a retrospective review of the unexpected infant deaths in the Southampton Health District and, together with the use of suitable controls, to evaluate the Sheffield at-birth scoring system or some modification of it with a view to implementing a risk-related intervention programme.

Before 1 January 1980 no special attention (in the sense of follow-up procedures now used routinely in Sheffield) was given to following up particular groups of babies in Southampton.

Materials and methods

The sudden unexpected infant deaths in Southampton are given as part of a confidential list published by the Hampshire Area Health Authority. This study covers those babies on that list aged between 1 week and 2 years of age dying suddenly

and unexpectedly between 1 July 1975 and 31 December 1979. All cases of sudden unexpected death were included even if a cause was later found at postmortem examination. Only sudden deaths of children with gross congenital abnormalities or malignancies were excluded. Our figures include some children who appear to have died in hospital rather than at home. These infants almost certainly died at home, although death was not certified until resuscitation in the accident and emergency department of Southampton General Hospital had been attempted.

For the determination of the Sheffield score the obstetric and perinatal histories of those children that could be traced to the birth registers of either the Southampton General Hospital or the Victoria House General Practitioner Maternity Unit were recorded. In addition, for each dead child two live controls were chosen. These were the children born immediately before and immediately after the index case, in the same maternity unit, and whose parents lived in Southampton at the time of birth. In the event of an index case being a twin the other twin was not used as a control.

Data on the eight relevant factors—maternal age, birthweight (g), number of previous pregnancies, duration of second stage of labour (min), mother's blood group, twinning, feeding intention, and presence of urinary tract infection during pregnancy—were obtained. Cases and controls were then scored using the “at birth” system.

Results

SUDDEN INFANT DEATH IN SOUTHAMPTON
The postperinatal mortality rate in Southampton for

1977–9 was 6·2 per 1000, slightly above the national average of 6·0 per 1000 for England and Wales. Table 1 shows the number of births, postperinatal deaths, and sudden infant deaths for the period 1 July 1975–31 December 1979 in Southampton. The seasonal distribution of the 68 sudden infant deaths is shown in the equi-areal rose diagram (fig 1) that takes into account the different numbers of days in different months, the leap year of 1976, and the six months of data in 1975.

Figure 1 shows clearly the excess of deaths in the winter months of December, January, and February as has been previously reported. A formal test for the absence of seasonality described by Mardia⁷ was significant ($\chi^2 = 13\cdot7$, $df = 2$, $p = 0\cdot001$) and suggested the peak incidence as the period around 1 January.

Table 1 Number of live births, postperinatal deaths, and unexpected infant deaths in Southampton, 1 July 1975–31 December 1979

Year	Live births	Postperinatal deaths		Unexpected infant deaths	
		No	Rate/1000	No	Rate/1000
1975	4802	43	9·0	8*	3·3
1976	4587	32	7·0	10	2·2
1977	4407	27	6·1	18	4·1
1978	4596	32	7·0	14	3·0
1979	4924	27	5·5	18	3·7
Mean/year	4663	32	6·9	15	3·3

*From 1 July to 31 December 1975 only.

Fig 1 Seasonal variation in sudden infant deaths.

EVALUATING THE SHEFFIELD SCORING SYSTEM

Material

The parents of 50 of the 68 babies who died suddenly (table 1) lived in Southampton at the time of birth. The maternity notes on all but three were obtained. The remaining 18 were not born in Southampton. Two additional cases were added, one who died on 30 June 1975 the other on 24 January 1980. This gave a total of 49 cases and their corresponding 98 controls.

Factors influencing "survival" times of dead babies

The age at death of the infants ranged from 18 to 434 days (median of 100 days). Figure 2 shows the survival curve of the 49 babies.

Of the eight factors included in the Sheffield at-birth score, only blood group appeared to effect the duration of survival among the index cases. Table 2 and fig 3 indicate an excess of early deaths among babies with maternal blood group O, and later deaths among those of blood group B.

There were 34 male and 15 female deaths.

Sheffield scores

Table 3 compares the index cases and controls from Southampton with those 195 cases and 250 controls used by Carpenter *et al*² with respect to each variable

Fig 2 Survival curve for babies classified as sudden infant deaths in Southampton 1975–9.

Table 2 Observed deaths by mother's blood group* and expected deaths assuming equal survival rates

Mother's blood group	No of deaths (O)	Expected deaths (E)	O/E	Prob
O	14	7·8	1·79	0·03
A	25	26·6	0·94	
B	9	13·6	0·66	

*The single case of an AB mother excluded.

Table 3 Comparison of cases and controls from Southampton and Sheffield

	Southampton		Sheffield	
	Cases	Controls	Cases	Controls
No of infants	49	98	195	250
Mother's age (years)	23.5	26.0	23.7	26.1
Previous pregnancies (mean)	1.7	1.2	1.8	1.3
% Caesarian section	12	7	3	6
Median duration of second stage of labour (min)	17.1	22.8	15.9	23.4
% Mother's blood group				
O	29	41	50	40
A	51	46	34	51
B	18	13	12	6
AB	2	0	4	3
Mean birthweight (g)	2981	3339	3042	3283
% Twin birth	10	0	6	4
% Feeding intention				
Breast only	41	65	15	29
% Urinary tract infection in pregnancy				
Absent	84	89	74	88
Query	8	6	14	7
Present	12	5	12	6

used in the Sheffield scoring system (table 4). The Southampton cases and controls appear similar to those from the Sheffield area with respect to mother's age, number of previous pregnancies, birthweight, and duration of second stage of labour. The proportion of mothers with blood group O in Southampton is 29% for the cases and 41% for the controls in contrast to 50% and 40% respectively in Sheffield.

The Sheffield scores for the 49 index and 98 control babies from Southampton were calculated in the standard way (table 4) and the resulting score distributions are shown in fig 4. The mean score for the index cases was 494, SD = 86 and for the controls 421, SD = 72.

It is clear from fig 4 that the proportion of cases (47%) with a score in excess of 500 is greater than the corresponding proportion for the controls (13%). A formal test of significance, incorporating the rest for

trend described by Armitage,⁸ is summarised in table 5 and is significant at $p < 0.001$, suggesting an increasing proportion of cases with increasing Sheffield score.

Survival analysis using the three score groups in table 5 when applied to the index cases suggests no association between score and time of death.

Table 4 Sheffield birth scoring system

Item	Score
Mother's age	$10 \times (50 - \text{age in years})$
Previous pregnancies	0
	1 21
	2 43
	3 64
	4 85
	5 107
	6 128
	7 149
	8 171
	9 or more 192
Duration of 2nd stage of labour	<5 min 127
	5-14 min 100
	15-29 min 72
	30 min-2 h 45
	>2 h 18
	NA 76
	Unknown 76
Mother's blood group	O, B, AB 44
	A 0
Birthweight (g)	<2000 93
	2000-2499 78
	2500-2999 62
	3000-3499 47
	3500-3999 31
	4000-4499 16
	4500-5500 0
Twin	Yes 103
	No 0
Feeding intention	Breast only 0
	Bottle or both 33
Urinary infection during pregnancy	Yes 54
	No 0
	? 5
Cut point for total score	500
	High risk 500 and over
	Low risk 499 and under

Fig 3 Survival curve for babies classified as sudden infant deaths in Southampton 1975-9 by mother's blood group.

Fig 4 Distribution of Southampton scores using Sheffield system, (49 cases, 98 matched controls).

Table 5 Proportion of Southampton cases in three score groups

	Sheffield score			Total
	≤399	400-499	≥500	
Cases	9	17	23	49
Controls	38	47	13	98
Total	47	64	36	147
Proportion of cases	0.19	0.27	0.64	

χ^2 Trend, 1 df = 16.76, $p < 0.001$.

Sheffield score modifications for Southampton

As there appear to be differences between the index cases and controls from Southampton and those of Sheffield with respect to some of the eight score variables summarised in table 3 it might be expected that using the method of discriminant analysis directly on the Southampton data would lead to improvement in separating cases from controls. Such a score system could be constructed, and one would expect the contribution of the variables to differ from that of the Sheffield system. The relevant analysis, however, is to determine whether the best predictor for the Southampton data represents a statistically significant improvement over the Sheffield score system.

The appropriate statistical theory described by Draper and Smith⁹ shows that the best approach is to calculate the Sheffield score S for each of the Southampton cases and controls and to use stepwise

discriminant analysis to investigate whether the discrimination achieved by S is statistically improved by adding in either singly or in combination any of the eight variables investigated.

In the event the analysis suggested that the only modification needed relates to mother's blood group. The problem of different associations among two case-control groups in Sheffield in this respect has been discussed by Carpenter *et al.*²

In view of the heterogeneity of the data sets in this respect the analysis was repeated excluding blood group from the Sheffield score. The resulting scores, S^* , could not be statistically significantly improved for the Southampton data either by any modification to the scores for the remaining seven variables or by including mother's blood group in some form. Reducing the cut point from 500 to 475 to allow for the omission of blood group S^* allocated 57% of the Southampton cases to the high risk group as compared with 15% of the controls.

Discussion

Suggestion that a scoring system of this type developed in a northern industrial city could be applied to a southern area with such a degree of apparent success is surprising, since each of the constituent factors of that system, blood group and twinning apart, may be heavily influenced by socioeconomic and environmental factors and since the scoring system did not appear to work in other areas, notably Birmingham.^{4,10} Evaluation of the Sheffield system has since taken place in nine other areas and has been found to be both effective and potentially useful with minor modifications.⁶ That the mother's blood group should be the only modification suggested for Southampton adds weight to those findings, especially since removal of the variable does not significantly alter the influences of the other factors.

Crucial to a study of this nature is the manner of selection of the cases. Our major criteria for choosing index cases were whether or not death had occurred at home, and then labelled as "cot death" after full pathological examination. Even allowing for variations in clinical and pathological practices it seems that most cases studied here could be grouped in the same way as those in the original Sheffield study,^{1,11} while only two of our 49 cases could be classed as accidental death. Of the other cases in Southampton 69% were group D: no disease; 12% group C: minor disease; and 16% group B: severe disease, compared with 33%, 37%, and 19% in the Sheffield study. Thus most cases in both studies were group C and D. No group A children (longstanding disease) were included in either study. A further

similarity of sex ratio, ages, and seasonal variations of the death between our cases and those in other studies confirms that we are studying typical cases of unexpected infant death.^{12 13}

The purpose of this study was to determine whether the criteria that existed could be used for detecting children at risk of cot death in Southampton and implemented within 48 hours of birth. The affirmative answer then raises the question: Is such a scoring system likely to be useful? Lewak *et al* constructed a remarkable system for identifying infants at risk based on eight factors.³ All eight risk factors were present in 56 of the 14 259 infants studied and six (11%) of these 56 infants died. Nevertheless, Lewak *et al* and more recently Valdes-Dapena¹⁴ concluded that "scoring systems are not yet sensitive enough for clinical use."

The only possible grounds for not using a birth scoring test is that one does not know what to do for the high risk infant. In which case the least one could do would be to give them a fortnightly check up, as has been done in Sheffield with encouraging results. Further, a theoretical evaluation of the Southampton system based on theory developed by Carpenter (unpublished observation) suggests that 40% of the postperinatal mortality might be eliminated by allocating the same total care in relation to risk. We are therefore planning to submit the birth system of scoring and risk-related intervention as developed in Sheffield to a prospective field study, attempting to direct the attention of the primary care team to the babies at greatest risk. It should not be overlooked that early intervention in pregnancy to improve the general level of antenatal care of the mother might significantly improve the state of the baby at birth and hence reduce the risk of sudden infant death.

Requests for reprints to: D Machin, Community Medicine, Southampton General Hospital, Tremona Road, Southampton SO9 4XY.

References

- ¹Protestos CD, Carpenter RG, McWeeny PM, Emery JL. Obstetric and perinatal histories of children who died unexpectedly (cot death). *Arch Dis Child* 1973; **48**: 835-41.
- ²Carpenter RG, Gardner A, McWeeny PM, Emery JL. Multistage scoring system for identifying infants at risk of unexpected death. *Arch Dis Child* 1977; **52**: 606-12.
- ³Lewak N, van den Berg BJ, Beckwith JB. Sudden infant death syndrome risk factors. *Clin Paediatr* 1979; **18**: 404-11.
- ⁴Oakley JR, Tavaré CJ, Stanton AN. Evaluation of the Sheffield system for identifying children at risk from unexpected death in infancy. *Arch Dis Child* 1978; **53**: 649-52.
- ⁵Palmer SR, Siggins RD, Bewley BR. Infant deaths in inner London: a health care planning team study. *Community Medicine* 1980; **2**: 102-8.
- ⁶Carpenter RG, Gardner A, Watson E. An epidemiological and sociological study of unexpected death in infancy in nine areas of Southern England. *Med Sci Law* 1981; **21**: 1-7.
- ⁷Mardia KV. *Statistics of directional data*. London: Academic Press, 1972; 357.
- ⁸Armitage P. *Statistical methods in medical research*. Oxford: Blackwell, 1974; 504.
- ⁹Draper NR, Smith H. *Applied regression analysis*. New York: Wiley, 1966; 407.
- ¹⁰Carpenter RG, Gardner A, Emery JL. Evaluation of the Sheffield system for identifying children at risk from unexpected death in infancy. *Arch Dis Child* 1979; **54**: 406-7.
- ¹¹Carpenter RG, Emery JL. Identification and follow-up of infants at risk of sudden death in infancy. *Nature* 1974; **250**: 729.
- ¹²Froggatt P, Lynas MA, Mackenzie G. Epidemiology of sudden unexpected deaths in infants (cot deaths) in Northern Ireland. *Br J Prev Soc Med* 1971; **25**: 119-34.
- ¹³Emery JL. Epidemiology of sudden unexpected or rapid deaths in children. *Br Med J* 1959; **ii**: 925-8.
- ¹⁴Valdes-Dapena MA. Sudden infant death syndrome: a review of the medical literature 1974-1979. *Paediatrics* 1980; **66**: 597-614.