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Evaluation of the Sheffield system for identifying children at risk from unexpected death in infancy

Results from Birmingham and Newcastle upon Tyne

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SUMMARY The 'at birth' system which is used in Sheffield to identify children likely to die unexpectedly in infancy, was tested retrospectively in Birmingham (83 cases) and in Newcastle upon Tyne (56 cases). The discrimination between cases and age-matched controls was poor in both cities. Analysis of the 8 factors used in the system showed that only 2 maintained significant case/control differences in Birmingham and Newcastle. Further investigation showed that other factors from maternity records showed significant case/control differences in these cities. Although the system used in Sheffield would not be of use in a prospective prevention programme in either Newcastle or Birmingham, the possibility of evolving an 'at risk' system which might apply more widely is discussed.

A system for identifying, early in life, children likely to die unexpectedly in infancy has been evolved in Sheffield (Carpenter *et al.*, 1977). Numerical weightings of 8 factors taken from obstetric and perinatal records allows nearly 60% of subsequent deaths to be identified in approximately 15% of the population, but the system may not be valid outside Sheffield. We report the results of applying the system retrospectively in Newcastle upon Tyne and in Birmingham.

Materials and methods

The obstetric and perinatal records were studied retrospectively for babies born in Newcastle upon Tyne (56 cases studied by A.N.S.) and in Birmingham (83 cases studied by J.R.O.). These babies had died unexpectedly between 1 January 1974 and 1 January 1977. All unexpected deaths in children aged between one week and 2 years were included, provided that there was no history of major congenital heart defect (diagnosed in life or discovered at necropsy) or neoplasia. Deaths from unnatural causes were also excluded. Those whom subsequent inquiry had shown to have had recognisable clinical evidence of acute illness, or an adequate pathological explanation for death, have been included, as have those who presented to hospital in a moribund state.

A living control was chosen for each case by taking the next live birth surviving from the same maternity hospital as the index, whose parents were living within the same city boundaries as the index parents at the time of birth.

Each case and control was 'scored' numerically according to the 'at birth' data analysis of Carpenter *et al.* (1977). The 8 factors used in this analysis, together with nearly 200 further variables obtained from the maternity and perinatal records, were subjected to a case/control comparison using a χ^2 test, with subsequent stepwise discriminant analysis. The probability value for each factor was obtained using the whole frequency of distribution of the data, and not from arbitrary groupings—except in the case of birthweight (500 g groupings) and maternal age (3-year groupings).

Results

Tables 1 and 2 show the degree of discrimination in each city between cases and controls, using the 'at birth' analysis of Carpenter *et al.* (1977). Overall, 72 (51·8%) of 139 index children and 42 (30·2%) of 139 control children scored 'at risk' ($P < 0{\cdot}025$).

There was no significant difference between cases and controls in either city for 6 of the 8 factors used in Sheffield (Table 3).

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Table 1 *Result of applying Sheffield scoring system retrospectively to 83 cases of unexpected death and age-matched controls in Birmingham*

At risk	*Controls*	*Indexes*	*Total*
Yes	27	38	65
No	56	45	101
Total	83	83	166

Table 2 *Result of applying Sheffield scoring system retrospectively to 56 cases of unexpected death and age-matched controls in Newcastle upon Tyne*

At risk	*Controls*	*Indexes*	*Total*
Yes	15	34	49
No	41	22	63
Total	56	56	112

For 3 of these factors (maternal ABO blood group, urinary tract infection during pregnancy, and length of second stage of labour) there were virtually identical distributions between indexes and controls. There was a tendency in the index populations of both cities towards mothers of higher parity, intention to bottle feed, and twins, as shown in Sheffield, but the differences between cases and controls were too small to achieve significance at the 10% level. A further discriminating factor in Sheffield—birthweight—maintained a significant difference between cases and controls in Birmingham only, although showing a nonsignificant trend towards low birthweight in index cases in Newcastle. Low maternal age was the only factor used in Sheffield which remained significant in both Newcastle and Birmingham.

Table 2 shows, for Birmingham and Newcastle, which of the factors from obstetric and perinatal records achieved differences between cases and controls that were significant at the 10% level or better. In each city significant differences were found in 8 variables using the χ^2 test, but using stepwise discriminant analysis 3 variables—admission to special care baby unit, number of invasive investigations to mother during pregnancy, and number of antenatal appointments kept by mother during pregnancy—were excluded from the Birmingham list.

In both cities, mothers of index children were younger, had a shorter interval between pregnancies, and 'booked' later at maternity hospitals, than mothers of control children. In Birmingham, index children were lighter at birth and regained birthweight more quickly than control children. In Newcastle the mothers of index children smoked more during pregnancy, attended more antenatal appointments, and received more parenteral iron than mothers of control children, while the index were of lower gestational age than control children and were predominantly male.

Discussion

The degree of discrimination that the Sheffield 'at birth' scoring system achieved in either Birmingham or Newcastle during the study period is not one that

Table 3 *Case/control comparison in Birmingham and Newcastle of variables used in Sheffield (marked *) and of other variables found to be significant at the 10% level or better (marked †) in Birmingham and Newcastle*

Variable	*Birmingham (n=83)*			*Newcastle (n=56)*		
	Cases	*Controls*	*Significance*	*Cases*	*Controls*	*Significance*
*Maternal age <24 years at delivery	46	38	†	49	43	†
*Birthweight <3000 g	40	26	†	20	13	NS
*Intending to breast feed	24	28	NS	12	19	NS
*Primiparous	22	32	NS	13	23	NS
*Twins	5	1	NS	2	0	NS
*Urinary tract infection during pregnancy	8	6	NS	7	7	NS
*Maternal blood group A	31	28	NS	20	24	NS
*Second stage of labour 10 min or less	29	24	NS	21	16	NS
Smoking during pregnancy	26	13	NS	30	13	†
More than 10 antenatal appointments kept at hospital	6	3	†	20	17	†
Invasive investigations during pregnancy	27	22	†	11	7	NS
Parenteral iron during pregnancy	1	0	NS	3	0	†
<3 years since birth of last child	42	20	†	34	15	†
Hospital booking before 20 weeks' gestation	41	55	†	27	44	†
Male infants	43	48	NS	39	26	†
Gestation 37 weeks or less	15	8	NS	13	6	†
Admission to special care baby unit	24	10	†	13	6	NS
Regained birthweight before discharge	40	24	†	23	22	NS

Arbitrary groupings are used to save space.
Probability values were derived from the frequency distribution of data.

would be of use in a prospective prevention programme, based upon the identification of a relatively large number of infants likely to die in a small proportion of the population. The sensitivity and specificity of the system were particularly low in Birmingham where fewer than half the indexes, but as many as one-third of the control population, were labelled 'at risk'.

Cases in Birmingham and Newcastle were selected in the same way as in Sheffield when the scoring system was established. The pattern of unexpected death in infancy has changed during the last decade in Sheffield (Forrest Hay and Emery, 1977), but the 'at birth' analysis is still sensitive for that community (J. R. Oakley, 1977, unpublished). We have no reason to believe that the reduced sensitivity of the system in Birmingham and Newcastle is owing to differences in causes of death in these cities.

Studies of the Oxford Record Linkage files (Fedrick, 1974a, b) showed no differences in maternal ABO blood group, length of second stage of labour, urinary infection during pregnancy, or feeding intention on discharge from hospital, between babies who died unexpectedly and controls. There was however, an association with twins, a shorter interval from the previous pregnancy, low maternal age, increased parity, and low birthweight.

Twins and increased parity have been found to be significant in previous studies of unexpected infant deaths (Valdes-Dapena, 1967). The failure of twins to discriminate in the present study may be due to the smaller number of deaths. By combining all 139 cases from both cities, 7 pairs of twins present in the index population, and one in the control population. Recent trends towards smaller families may explain the reduced association between high parity and unexplained death.

The mother's intention to breast feed her baby is used in the 'at birth' analysis, but this may not be a good indication of the subsequent feeding pattern. Analysis of feeding pattern after discharge *may* substantiate an association between artificial feeding and unexpected death, but the use of modified milks may have reduced its importance.

We believe that the association of maternal ABO blood group, urinary infection during pregnancy, and the length of second stage of labour with unexpected death in infancy in Sheffield may be chance findings, as the distribution of each of these factors was virtually identical between cases and controls in both Birmingham and Newcastle. Some of the associations in these 2 cities may be due to chance, and we do not claim that the factors found to be significant in Newcastle or Birmingham have any more importance than those in Sheffield.

It would be possible to improve the degree of discrimination in Birmingham and Newcastle by replacing some of the factors used in Sheffield with ones that were more valid in each city. This would only be a local solution, because some of the factors would not be applicable to other communities. If a scoring system is needed to improve infant mortality rates in this country, one should be devised which could apply to any community. This would mean using factors that maintain similar differences between cases and controls in many communities. Our data suggest that sufficient numbers of such factors may not be available from obstetric and perinatal records.

Additional data on each child and his environment can be collected after he has gone home. The second stage of the scoring system suggested in Sheffield includes an assessment of the home and the difficulties in establishing feeding. Such factors are based on the subjective evaluation of the observer, and would be difficult to apply to different communities using many observers. It may be helpful to use simple geographical data, as previously reported (Oakley and Tavaré, 1977). Such data were also found to be important in Nottingham (Madeley, 1977).

A DHSS multicentre postneonatal study of 11 areas, with a combined population of over five million people, has been in progress since April 1976. It remains to be seen whether data from this study will enable a better discriminant to be devised using objective and reproducible environmental and social factors to augment the hospital records. This discriminant may help to plan a wider prospective programme to prevent unexpected deaths in infancy. At present, any area wishing to establish its own system to identify those at risk is likely to have too few recent cases for statistical analysis. Almost certainly there will be too few deaths to determine if identification can lead to prevention, or even to tell if identification is an acceptable alternative to more and better care being given to *all* infants during their first few months of life.

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