Sudden infant death syndrome in south east Scotland

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SUMMARY Three hundred and fifty eight infants from south east Scotland who died suddenly were classified into four groups. Categories for these groups ranged from where a definite cause of death had been recorded to where no explanation had been provided and no associated disorder was discovered (SIDS). Our results supported the view that there are few differences in the history of cases certified as SIDS and other cases reported as dying suddenly but with an explanation. Groups that most closely matched the SIDS definition employed were reported to be healthier throughout life and freer from illness in the 48 hours before death. From the findings of this study the 'true' SIDS group did not appear as an 'at risk' population. The study group as a whole was not marked by social deprivation, poor mothering, or less privileged families. The importance of intensive investigation, including postmortem examination was emphasised, as misdiagnosis may give a 'falsely' inflated picture of the incidence of the syndrome and could cause unnecessary anxiety.

A definition commonly employed in the identification of instances of the sudden infant death syndrome (SIDS) is that proposed in Seattle in 1969: "The sudden death of any infant or young child which is unexpected by history, and in which a thorough post-mortem examination fails to reveal an adequate cause of death."¹

In this study data from a group of infants who died suddenly and unexpectedly and who presented as cases of SIDS were analysed to determine to what extent they matched the above definition and to study any differences detected.

The criterion for the diagnosis of SIDS was necessarily pathological as each infant's death was unexpected by history. To carry out this investigation and enable discrimination between the pathological findings, four categories of classification were employed. These classifications, A, AB, B, and C, enabled analysis of the various findings and study of the extent to which each may have contributed to the death.

The variables used as parameters for comparison between the groups were selected on the basis of their citation in reviews that dealt with SIDS where possible risk factors, such as preterm delivery, mother's age less than 20 years, and poor social conditions surrounding the family, have been indicated.^{2,3}

Procedure

From 1977 to 1985, 358 infants aged between 1 week and 2 years who died suddenly and unexpectedly in south east Scotland were referred by the procurator fiscal for postmortem examination. These infants, all of whom presented as cases of SIDS, represented 80% of fiscal referrals. The 20%, that were excluded failed to meet fully the definition of SIDS given above.

Each case underwent radiological, bacteriological, and virological studies. Biochemical estimates of the sodium, potassium, and urea concentrations in the vitreous humour of the eye were obtained and a detailed postmortem examination performed by a paediatric pathologist. Data on each case were abstracted from obstetric and paediatric records about the pregnancy and birth and any further admissions for the infant. An extensive protocol was used that covered the mothers' antenatal history, parity, interval to last live birth, and regularity of clinic attendances. Problems during pregnancy, such as hypertension, urinary infection, anaemia, and admissions to hospital, were noted.

For the infant, data about mode of birth, gestational age, birth weight, and admissions to the special care baby unit were recorded. Subsequent admissions to hospital were noted by cause, duration of stay, and interval from discharge to death.

Each family doctor was sent a proforma for completion, which dealt with the mother's previous medical and social history and provided for details regarding any problems with the infant and the family. The health visitors also received a form on which to record their knowledge of the family and infant history, including attendance at the health clinic and visits to the home.

A letter was written to each family with the offer of a visit to the home. This visit was undertaken in every case by the same experienced interviewer two to three weeks after the death when a full history was taken about the infant's health, feeding patterns, circumstances surrounding the death, and household and social organisation of the family. An estimate was made of the reliability and facility of the parents' recall. In 27 cases (7%) a visit was not carried out as the families had either left the area or intimated their wish not to be interviewed.

Classification into one of four groups was carried out after the interviews and investigations had been completed to reduce the possibility of bias on the part of the investigators.

Group A (26 cases) comprised those cases where a definite cause of death was ascertained. Group AB (53 cases) comprised those cases where an associated disorder was detected that may have been a factor in the death. Group B (193 cases) comprised those cases where an associated disorder was present but was not thought to be an adequate cause of death. Group C (86 cases) comprised those cases where no observable associated disorder was present.

Analysis of the data relating to each variable was carried out to determine whether or not significant differences existed between the groups. Statistical procedures were based on Freund and Scheffé.^{4,5}

The classification of cases into group A was made where an adequate cause of death was found. These deaths, though presenting as SIDS, could no longer be certified as such as they did not completely match the definition. Causes of death in this group were infections (10 cases), Reye's syndrome (five), congenital cardiac defects (four), myocarditis (two) and one each of Waterhouse-Friderichsen syndrome, malnutrition, hypernatraemic dehydration, laryngeal cysts, and a ribbon impacted in the larynx.

The inclusion of cases into group AB relied on the judgment of the pathologist about the severity of the disease process present and the extent of its contribution to death. This was generally assessed histologically. As stated in one report, 'there can be doubt and ambiguity, even after a thorough paediatric post-mortem, as to what constitutes an adequate cause of death'.⁶ Most postmortem examinations in this study were undertaken by the

same prosector, which gave uniformity to both the performance and interpretation of the examination and contributed to the reliability of the findings. A pathogen was isolated and histological evidence of infection shown in 39 cases (73.6%) in this group. In the other 14 cases (26.4%) a variety of other diagnoses were found, such as a degree of nephrocalcinosis, inhalation of vomit, or metabolic disorders, but in no instance was the condition thought to be severe enough to be an adequate explanation for cause of death.

In most cases in group B there was a positive bacteriological or virological isolation but with no histological evidence of even minor infection. The classification B was used to show the presence of some positive finding, thus distinguishing the group from group C, which had no positive finding.

Results

General factors. The greatest difference between any two groups in the proportion of homes that had no visit was found between groups A and AB, but the difference was not significant ($p=0.197$, Fisher's test of exact probability).

The social status of the family was determined by the occupation of the father, using the Office of Population Censuses and Surveys' Classification of Occupations.⁷ The only significant result obtained was that produced by a χ^2 test applied to groups B and C ($\chi^2=7.29$, $df=2$, $p<0.05$). The most pronounced differences were in social classes one and two (19% of group B and 34% of group C) and in social class three (44% of group B and 34% of group C).

The family doctor, health visitor, and interviewer made an independent assessment of each family as regards parental supervision and management of the infant's feeding. The state of repair of the house and furnishings were also evaluated. Analysis of the gradings (good, average, or poor) produced a non-significant value for F of 1.57 ($df=3$ and 342). Almost all estimates (98%) were average or good.

Maternal factors. Analysis of the mothers' ages at the birth of the infants showed no significant differences between groups ($F=2.90$, $df=3$ and 353). There were also no significant differences between groups when mothers aged under 20 were compared ($\chi^2=2.00$, $df=3$), the percentage of these young mothers in the groups ranging from 9% in group AB to 19% in group A.

Over half (53%) of the mothers smoked. The highest percentage of smokers was in group AB (58%) and the lowest in group C (50%), but the differences were not significant ($\chi^2=0.79$, $df=3$).

Comparison of small for gestational age births⁸ in each group among smoking and non-smoking mothers produced a non-significant result ($\chi^2=5.22$, $df=3$). Similarly, no significant difference was found when the infant was both small for gestational age and below the 10th centile at death ($\chi^2=0.10$, $df=1$).

After the first born infants had been omitted the number born more or less than 16 months since the mother's last live birth were compared between groups. The only significant result was that 37% of group A had an interval of less than 16 months compared with 14% of group B ($\chi^2=5.77$, $df=1$, $p<0.05$).

Most mothers (76.3%) had booked with their own doctor or at the maternity hospital by 12 weeks' gestation, with the highest percentage not booking by this time in group A (33%) and the lowest in group C (18%). The differences in booking times were not significant ($\chi^2=3.976$, $df=3$).

Differences between the groups were small and non-significant ($\chi^2=0.32$, $df=3$) when antenatal complications were analysed, complications being present in 51% of group AB, 48% of group C, 48% of group B, and 46% of group A.

Infant factors. Group A contained the highest percentage (38%) of first born infants compared with 19% in group AB and 27% in group C. Proportions of second born infants were similar in groups AB, B, and C (57, 46, and 44%, respectively), with the lowest representation in A (31%). The highest representation of boys was in group C (66%) and the lowest in group A (50%), but these differences were not significant ($\chi^2=2.56$, $df=3$).

The mean duration of gestation for the four groups ranged from 38.3 weeks (group A) to 38.9 weeks (group B), a non-significant difference by analysis of variance ($F=1.39$, $df=3$ and 354).

The mean birth weights ranged from 3154 g (group B) to 3052 g (group AB), but the difference was not significant by analysis of variance ($F=0.32$, $df=3$ and 349). Birth weights ≤ 1500 g represented from 1.1% of group B to 3.8% of group A. The largest number of small for gestational age infants was in group AB (23%) and the smallest in group C (13%). The difference was not significant ($\chi^2=3.64$, $df=3$).

Differences for mean (SD) crown to heel length were small and not significant (analysis of variance, $F=0.34$, $df=3$ and 288), the shortest being 49.65, (3.43) cm (group AB) and the longest 50.40 (3.43) cm (group A).

Infant's health. Thirty per cent of group AB, 16% of group B, 15% of group C, and 12% of group A were

admitted to the special baby unit some time after birth. Differences were not significant ($\chi^2=7.168$, $df=3$). Fewer infants in groups A and AB (15% and 4%, respectively) were reported as healthy than in groups B and C (21% in both groups), the differences reaching significance ($\chi^2=9.17$, $df=3$, $p<0.05$).

Infants in group A were admitted to hospital twice as much as infants in groups B and C, but this was not significant ($\chi^2=6.43$, $df=3$).

When groups A and AB were compared with groups B and C about the health of the infants 48 hours before death significant differences were obtained. The percentage (69%) of group A who were not healthy was significantly higher than group B (40%) ($\chi^2=8.03$, $df=1$, $p<0.01$) and group C (30%) ($\chi^2=12.73$, $df=1$, $p<0.001$). Similarly, the percentage of group AB who were not healthy (60%) was significantly higher than group B ($\chi^2=7.07$, $df=1$, $p<0.001$) and group C ($\chi^2=12.25$, $df=1$, $p<0.001$).

Factors at death. Mean (SD) age at death was lowest in group C (16.3 (13.2) weeks), highest in group AB (25.3 (20.9) weeks), and for group A was 22.0 (20.8) weeks and for group B was 18.1, (114.0) weeks). As there were significant differences between these means when analysed by an F test ($F=4.28$, $df=3$ and 354, $p<0.05$) a Scheffé test was employed to determine where these differences were. At death group AB was significantly older than both group B and group C ($p<0.05$ in both cases) and group A was significantly older than group B ($p<0.01$).

More infants died in the coldest months (40% in December, January, and February). The number of deaths in these months within the groups ranged from 46% in group A to 34% in group C, but these differences were not significant ($\chi^2=1.99$, $df=6$).

The type of feeding at time of death showed a range for breast feeding alone from 31% (group A) to 13% (group AB) and for bottle feeding alone from 62% (group B) to 49% (group C). Significantly more infants were bottle fed in group B than in group C ($\chi^2=4.01$, $df=1$, $p<0.05$).

The mean interval from finding the infant dead to the time of postmortem examination for all cases was 38.9 hours. Infants below the 10th centile on weight for age tables employed in hospitals where the postmortem examinations were performed comprised 52% of group A, 42% of group AB, 26% of group B, and 29% of group C. The difference between the groups was significant ($\chi^2=9.93$, $df=3$, $p<0.05$).

For the crown to heel measure, allowance was made for age by constructing T scores based on local

norms for each age group. A non-significant result ($F=1.56$, $df=3$ and 272) was obtained.

Discussion

Previous studies have shown that there are few, if any, differences between cases certified as sudden infant death syndrome and other cases reported to the coroner or procurator fiscal as unexpected deaths.⁹⁻¹³ Our study, which has found few significant differences between the groups based on degree of information, agrees with this.

Some features commonly associated with sudden infant death syndrome—namely, temporal variation, age distribution, and slight male predominance—were seen in our groups, but in contrast with most previous studies, there was no over representation of SIDS among families towards the lower end of the social class scale. The group as a whole was not marked by social deprivation, poor mothering, or less privileged families.

All infants in the study were reported as dying suddenly and unexpectedly. Classification took place only after postmortem examination and histological analysis. The present data indicated non-pathological differences between the groups, some of which may have been predicted, perhaps being an artefact of the criteria employed in classification—for example, the pattern of incidence of reported ill health just before death in groups A and AB.

Group A, because death was sudden and unexpected by history, could only match the adopted definition in that respect as a cause of death was found. For this reason they could not be classified as SIDS. This group produced the highest proportion of the main epidemiological factors associated with SIDS, thus supporting the finding that these variables are also associated with an increased incidence of all infant deaths.¹⁴

Most of the evidence for the influence of those variables commonly associated with increased risk of sudden infant death was found in groups A and AB (Fig. 1). It has been well documented that children with conditions such as hypernatraemic dehydration and malnutrition are liable to come from homes where there is a considerable amount of substandard care,^{6 15 16} but as there were only two such cases in our study it was considered that these did not introduce a selection bias.

These two groups (A and AB) also included a comparatively high representation of infants whose weight at postmortem examination fell below the 10th centile (Fig. 2). This may be partly due to postmortem dehydration, but as there was no longer or shorter death/necropsy interval between all the groups the presence of an underlying disease process

may account for this finding. It has been suggested that growth patterns of some SIDS victims may be similar to those attributed to smoking during pregnancy.¹⁷ Our data, which showed no significant difference between groups as regards maternal smoking, small for gestational age infants, and those who fell below the 10th centile, do not support this theory.

For purposes of certification of death in group AB it may be more practicable to adopt the practice used in Sheffield, in which double registration enables a disease process to be identified and the death also to be described as SIDS.¹⁸ Like a previous study,¹⁹ we had reservations about including these subjects (group AB) in a 'true' SIDS group. Because of the element of doubt we decided, for the sake of the validity of the study, to form a separate group for these cases.

This group would match most closely 'Category B' as defined by Taylor and Emery,¹⁸ where factors

Fig. 1 Influence of variables commonly associated with increased risk of sudden infant death in the four groups (group A=cases where a definite cause of death was ascertained ($n=26$); group AB=cases where an associated disorder was detected that may have been implicated ($n=53$); group B=cases where an associated disorder was detected but not implicated ($n=193$); group C=cases where no observable associated disorder was present ($n=86$)). Variables, reading left to right, are: Mother <20 years, socioeconomic state (SES), gestation <37 weeks (Gestation), booking time (at hospital) <3 months (Booking), last live birth <16 months ago (Last LB <16), infant not healthy, weight at postmortem examination <10th centile (Weight <10th), number of admissions to hospital (HA), and admissions to special care baby unit (SCBU). Values are percentage of those in group with that variable.

Fig. 2 Percentages of infants from the four groups (group A=cases where a definite cause of death was ascertained ($n=26$); group AB=cases where an associated disorder was detected that may have been implicated ($n=53$); group B=cases where an associated disorder was detected but not implicated ($n=193$); group C=cases where no observable associated disorder was present ($n=86$)) who were small for gestational age at birth and whose weight at postmortem examination fell below the 10th centile.

were found that were 'possibly treatable'. Death may have been preventable in some cases in group AB where it was possibly the result of an inappropriate response to infant illness by the caregiver or medical attendant.⁶ The inclusion and identification of a group 'AB' of this nature in SIDS studies may contribute to understanding the role of conventional paediatric treatment in the reduction of infant mortality.

The finding that there were less bottle fed infants in group C than in group B probably reflected social class, significantly more families from social classes I and II being in group C. These two groups were similar for most measures. Some factors commonly associated with SIDS, however, were apparent (second borns, age at death, and winter peaks) and in agreement with the findings in another study²⁰ support for most attributed influences, such as high incidence of mothers aged less than 20, short inter-pregnancy intervals, and premature birth, was not forthcoming. From these results it may be concluded that these 'true' SIDS infants would not appear as an 'at risk' population.

In Sheffield, where attempts have been made to identify infants at risk from unexpected death, the proportion of high and low risk infants varied considerably in different 'causal groups'.²¹ Comparison of this study with ours is difficult due to differences in classification, but there was a similar absence of 'risk' factors in the unexplained groups.

Compared with other infant deaths in Scotland, deaths due to SIDS are not significantly different when analysed by sex of child, social class of father, and age of mother.²² Our study supports Sunderland's view that all infants who die suddenly and unexpectedly are not always cases of SIDS.²³ Many published reports on SIDS are confused by variation in definition. The term 'cot death' is often employed rather than SIDS but in fact only refers to the mode of death—that is, dying unexpectedly and suddenly.¹³

Bergman's definition¹ was created to encompass those deaths that were sudden and unexpected and in which adequate cause had not been established. To date, we believe that this definition has not been bettered, even though it may be reconsidered as recommended in a recent report⁶ as it does not describe consistently the degree to which the death was unexplained.

The diagnosis of SIDS is one of exclusion of recognisable causes of death. Perhaps it should also include the consideration of adverse circumstances surrounding the death in the manner described by Bass *et al*, where fuller investigations into the circumstances surrounding death in 24 cases of certified SIDS changed the diagnosis in all instances.²⁴ In the present study the circumstances surrounding death were fully investigated, by the police, before postmortem examination, and it is considered that the classification employed has permitted a group of 'true' SIDS to be identified (groups B and C), a group that also rigorously matched the standard definition of the syndrome set out above. The importance of a postmortem examination with prior and subsequent investigations provides a more reliable estimate of the incidence of SIDS. In areas where diagnosis is only made clinically—that is, where there is no autopsy—there may be over reporting or under reporting of the syndrome. An accurate diagnosis is beneficial in counselling bereaved parents and in the management of families who have or wish to have subsequent children, particularly about the question of the recurrence risk for SIDS and the use of apnoea monitors.

The diagnosis of SIDS may too often provide an 'umbrella' to cover cases where although death may have been sudden and unexpected, it was not always totally unexplained.

In studies of sudden infant death there is often concern about the question of infanticide. The Infanticide Act does not apply in Scotland. Were there to be a charge it would almost certainly be that of culpable homicide.²⁵ In the present study only two instances had some suspicion of parental mis-handling.

'True' SIDS remains the largest single cause of death in infants who die suddenly and unexpectedly. Until a diagnostic marker or markers have been identified and agreed on, the diagnosis of SIDS remains dependent on the judgment of the pathologist. Misdiagnosis serves to confuse those seeking the cause(s) of these tragic deaths. This may give a falsely inflated picture of the incidence of the syndrome and also produce an unnecessarily high level of anxiety among parents.

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