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Hazardous cosleeping environments and risk factors amenable to change: case-control study of SIDS in south west England

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ABSTRACT

Objectives To investigate the factors associated with sudden infant death syndrome (SIDS) from birth to age 2 years, whether recent advice has been followed, whether any new risk factors have emerged, and the specific circumstances in which SIDS occurs while cosleeping (infant sharing the same bed or sofa with an adult or child).

Design Four year population based case-control study. Parents were interviewed shortly after the death or after the reference sleep (within 24 hours) of the two control groups.

Setting South west region of England (population 4.9 million, 184 800 births).

Participants 80 SIDS infants and two control groups weighted for age and time of reference sleep: 87 randomly selected controls and 82 controls at high risk of SIDS (young, socially deprived, multiparous mothers who smoked).

Results The median age at death (66 days) was more than three weeks less than in a study in the same region a decade earlier. Of the SIDS infants, 54% died while cosleeping compared with 20% among both control groups. Much of this excess may be explained by a significant multivariable interaction between cosleeping and recent parental use of alcohol or drugs (31% v 3% random controls) and the increased proportion of SIDS infants who had coslept on a sofa (17% v 1%). One fifth of SIDS infants used a pillow for the last sleep (21% v 3%) and one quarter were swaddled (24% v 6%). More mothers of SIDS infants than random control infants smoked during pregnancy (60% v 14%), whereas one quarter of the SIDS infants were preterm (26% v 5%) or were in fair or poor health for the last sleep (28% v 6%). All of these differences were significant in the multivariable analysis regardless of which control group was used for comparison. The significance of covering the infant's head, postnatal exposure to tobacco smoke, dummy use, and sleeping in the side position has diminished although a significant proportion of SIDS infants were still found prone (29% v 10%).

Conclusions Many of the SIDS infants had coslept in a hazardous environment. The major influences on risk, regardless of markers for socioeconomic deprivation, are amenable to change and specific advice needs to be given, particularly on use of alcohol or drugs before cosleeping and cosleeping on a sofa.

INTRODUCTION

The decrease in numbers of deaths from sudden infant death syndrome (SIDS) after the "Back to Sleep" campaign in the early 1990s has been followed by a slow but steady reduction of almost 50% since the late 1990s.¹ However, it is not clear whether the subsequent fall in SIDS rates results from avoiding placing infants in the prone position to sleep or the uptake of other more recent advice.

The original risk reduction messages in 1991 included avoiding placing infants in a prone sleeping position, reducing the amount of bedding, and recommending that mothers stop smoking during pregnancy. During 1993-6, as part of the Confidential Enquiry into Stillbirths and Deaths in Infancy, we carried out a national study to identify further risk factors for SIDS, including placing infants in a side sleeping position, cosleeping (particularly with a smoker), infants sleeping in a room alone, use of duvets and pillows, whether bedclothes covered the infant's head, and postnatal exposure to tobacco smoke. Subsequent national advice was to avoid each of these factors and place infants in the "feet to foot" position—that is, with their feet at the foot of the cot.²⁻⁴

Relatively little is known about the uptake or distribution of these messages in different social or cultural groups, the relative importance of changes in known epidemiological risk factors for SIDS, or the emergence of new or previously unrecognised factors.

Recent studies of the residual deaths also show a high proportion of infants cosleeping.⁵⁻⁸ The proportional increase in deaths from SIDS among infants cosleeping has led some authorities, including the American Academy of Pediatrics,⁹ to recommend against bed sharing. Using longitudinal data in Avon over a

20 year period, however, we found that this proportional increase was due to the noticeable reduction in SIDS deaths occurring in the cot environment rather than a numerical increase in SIDS deaths while the infant coslept in the parents' bed.¹⁰ Findings from our national study 10 years ago also suggested that the risk to the infant might be due to the circumstances in which cosleeping occurred rather than cosleeping itself.¹¹ Thus one of the primary hypotheses of this investigation was to look at the type of surfaces on which the adults and infants coslept and any interaction between cosleeping and recent parental alcohol or drug use.

We report a four year population based case-control study of all unexpected infant deaths from birth to age 2 years in the south west of England, 10 years after our national study.² The present study includes an investigation of the scene of death and the scene of sleep in two groups of controls.¹² To investigate whether some of the risk factors might merely be markers of socio-economic deprivation, we included a high risk control group weighted to several epidemiological risk factors for SIDS, as well as a randomly selected control group.

METHODS

From January 2003 to December 2006 we carried out a population based case-control study of all sudden unexpected deaths in infancy in the counties of Gloucestershire, Wiltshire, Bristol, Somerset, Devon, and Cornwall, in the south west of England. We aimed to include all sudden unexpected deaths from birth to age 2 years in a total study population of 4.9 million. The age range extended beyond the usual 12 month cut-off as it is recognised that, although rare, unexpected deaths in the second year of life share many characteristics with those in infancy. Where possible we followed the guidelines for strengthening the reporting of observational studies in epidemiology.¹³

To ensure that all sudden unexpected deaths were notified within 24 hours, the established notification network system used in Bristol (including ambulance control centres, accident and emergency departments, police, coroners' offices, paediatricians, parents' groups, and health centres) was extended to the whole of the study region using a dedicated telephone line that was checked twice a day.

The full multiagency protocol^{12,14} for care of bereaved families and investigation of the circumstances of the death was initiated immediately after notification. A full history, including a narrative account from the parents, was collected as soon as possible and a home visit including an investigation of the scene and circumstances of death was carried out by a trained paediatrician or health visitor together with a member of the police child protection team. Provision of clinical and bereavement care to the families was not conditional on their consent to take part in the study. Consent for inclusion in the study was sought several days after the death, and families who consented were visited by one of the research team within two weeks to complete a further detailed questionnaire. The cause of

death, using the Avon clinicopathological classification,¹² was established at a multidisciplinary review meeting at which all available information and records were reviewed, including the results of a full paediatric autopsy to a standard protocol.^{2,14} We focus on those unexpected infant deaths (up to age 2 years) that remained unexplained at the end of the multiagency investigation and thus met the definition of SIDS.^{12,14}

We identified two groups of control infants at 28-30 weeks' gestation from the stork maternity database. These infants were followed up longitudinally using five sequential postal questionnaires to eight months after birth: a randomised control group and a group identified as being at high risk of SIDS. The high risk group was chosen using the most significant prenatal predictors of SIDS modelled from a previous study,² which included maternal smoking, larger families, younger mothers, and mothers classified as social class IV (partly skilled), V (unskilled), or never employed using the registrar general's occupational coding. The database at St Michael's Hospital in Bristol includes about 450 births each month, and the two groups of control families were selected each month over a period of two years; we aimed to enlist 300 randomly chosen families and 150 high risk families. The initial request to participate in the study was by a letter delivered by the community midwife allocated to each mother. Because of this indirect contact, as requested by the ethics committee, we anticipated a relatively low initial uptake, particularly among the more socially disadvantaged families. To minimise potential selection bias in the randomly selected controls we weighted the selection process for "random" controls to match the maternal social class distribution of mothers with dependent children in Avon from the 1991 census. The risk of selection bias among the high risk controls was thought to be negligible as the families selected for inclusion were some of the most deprived and thus the type of families that are often under-represented in most observational studies.

We aimed to interview 80-100 families from each group at home using the same questionnaire administered to the families that experienced sudden unexpected deaths in infancy, and identified a specific time of sleep as reference within 24 hours of the interview. The age of the infants at interview and the time of day of the reference sleep were weighted to reflect approximately the ages and times of day at which infants had died unexpectedly.

Definition of variables

Cosleeping was defined as an infant sharing the same bed or sofa with an adult or child. We collected information on both prescribed and illegal drugs used by the parents or carers, medicinally or for recreation. The alcohol limit of no more than two units was based on recent UK recommendations of the maximum daily intake for women. To assess the interaction between cosleeping and alcohol or drug use both the questionnaire responses and the data given in the

narrative account of the parents were checked to ensure that the parent who had consumed these substances was the same parent who had coslept with the infant.

Although some of the information collected was sensitive, especially on use of alcohol and drugs, we are confident that our use of specialist health visitors and emphasis on study confidentiality enabled us to obtain a sufficiently accurate account of what happened.

Statistical analysis

We used medians and interquartile ranges to describe data that were not normally distributed. For categorised continuous variables where a commonly used cut-off was unavailable we used the data below the 25th percentile from the combined dataset. Odds ratios, 95% confidence intervals, and P values for the univariable and multivariable analysis were calculated using logistic regression in SPSS. As the weighting of factors is not an exact process we adjusted all comparisons with the random controls for infant age and whether the final sleep was during the day or night by including these factors in the model regardless of their significance; comparisons with the high risk controls were further adjusted for maternal smoking during pregnancy, maternal age, number of live births, and maternal social class. To construct models we used the backward stepwise procedure for variables significant at the 5% level in the univariable analysis. At the end of the modelling process we tested any variables with more than 5% of data missing among the cases and controls.

RESULTS

During the four years of the study period 157 apparent sudden unexpected deaths in infancy occurred in a population of 184 800 births. Two of these deaths were subsequently found to have occurred in infants with known life threatening conditions, leaving 155 sudden unexpected deaths in infancy. A review of alternative sources of information on infant deaths (including returns to the Confidential Enquiry into Maternal and Child Health, the local press, communication with coroners, and contact with paediatricians and police) identified a further two unexpected deaths that met the inclusion criteria but were not notified to us directly. Thus in the study area within the research period we were notified of 155 of 157 (99%) sudden unexpected deaths in infancy. Of these deaths a causal explanation was established for 67 (43%), and 90 (57%) were classified as SIDS, which equates to a rate per 1000 live births of 0.49 (95% confidence interval 0.40 to 0.60), a rate similar to that in England and Wales during the study period. Of the 90 infant deaths classified as SIDS, 10 were excluded according to the study protocol: four families did not give consent, three deaths occurred before approval was obtained from the local ethics committees, two families moved away, and one death was initially suspected as an intentional injury but not confirmed. Thus 80 of 90 (89%) deaths due to SIDS were included for analysis.

Detailed findings from the longitudinal questionnaire data are to be reported in detail elsewhere. Overall, 385 randomly chosen control families and 206 high risk control families agreed to take part in the study and completed the first questionnaire during pregnancy. Over the 24 months of the recruitment period we contacted 1438 random control families and 1191 high risk control families to ensure that our targets were met, and we used a weighting strategy to offset selection bias. When the control infants reached a specific age we approached 92 random control families and 95 high risk control families for interview. We include the data collected from the 87 random control families (95%) and 82 high risk control families (86%) who were interviewed at home. The data on infant care practices and sleep environment thus relate to the night or day that the infants died from SIDS (n=80) and to the reference sleep within the 24 hours before the interview for the two control groups.

Weighting of controls

Weighting the randomly chosen families to match maternal socioeconomic status with census data was largely successful (fig 1). One third of the randomly chosen mothers were social classes IV (partly skilled), V (unskilled), or never employed compared with a similar proportion of the Avon mothers (34% v 33%). The census data of mothers with dependent children in the south west region show a similar distribution.¹⁵

Table 1 compares the weighting factors between the cases and controls. Infant age and time of sleep were comparable in all three groups, 84% of SIDS infants died during what the parents considered to be night time sleep. With the weighting process the high risk control families reflected characteristics closer to those of the families with SIDS infants than the randomly chosen counterparts: younger mothers, mothers more likely to smoke, larger families, and more social deprivation. This was also reflected in factors not used in the weighting process, to measure the poorest end of the social spectrum: including families

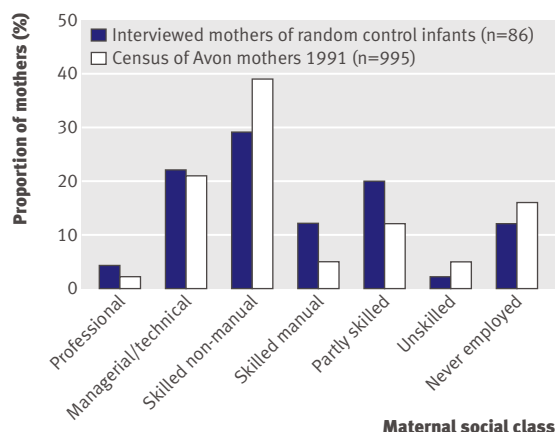


Fig 1 Occupation of random control mothers compared with 1991 census data on mothers with dependent children from Avon

Table 1 Weighting factors used between SIDS (sudden infant death syndrome) infants and random or high risk control infants. Values are numbers (percentages) unless stated otherwise

Factor	SIDS infants	Random control infants	High risk control infants
Median (interquartile range) infant age (days)	67 (39-144)	69 (42-126)	68 (28-145)
Died during night time sleep (as defined by parents)	67/80 (84)	67/87 (77)	65/82 (79)
Maternal social classes IV, V, or never employed*	52/77 (68)	29/87 (33)†	50/80 (63)
Median (interquartile range) maternal age (years)	24 (21-30)	29 (24-34)†	25 (21-32)
Maternal smoking during pregnancy	47/79 (59)	12/87 (14)†	36/82 (44)
≥3 live births	32/80 (40)	17/87 (20)†	27/82 (33)

*IV=partly skilled; V=unskilled.
†Non-weighted factors.

with a weekly income of less than £100 (€114; \$167; SIDS 16%, random controls 5%, high risk controls 11%) and living in houses with serious damp and mould (SIDS 14%, random controls 1%, high risk controls 7%). The weighting factors were further adjusted for as appropriate in subsequent univariable and multivariable estimates.

Figure 2 shows the age distribution for the SIDS infants and the ages at which home visits were carried out for the random and high risk control infants. The distribution of ages at which home visits were carried out for the control groups was similar. The age distribution of the SIDS infants was significantly different from previous studies, with a modal value between 5 and 8 weeks and a median of 66 days, interquartile range 39-130 days); this was more than three weeks younger than in the Confidential Enquiry into Stillbirths and Deaths in Infancy (median 91, 55-150 days).

The seasonal distribution of the SIDS deaths was uniform and thus reflected in the timing of interview for the reference sleep among the controls. Deaths from SIDS were more common among males (51/80, 63.8%), which was also the case among both sets of controls, yielding no significant difference by sex.

SIDS infants v random control infants

Significant multivariable findings

Table 2 lists those factors that were significant in the univariable analysis and the adjusted risk for those factors that remained significant in the multivariable regression model. The strongest single factor in this model was maternal alcohol consumption. Of the SIDS infants, 16% (13/79) had been cosleeping on a sofa but because only one control infant had slept in a similar environment we combined those infants who shared their parents' bed and those who shared a sofa into a single group (despite the noticeable difference in risk between the two environments). Overall, 54% of SIDS infants had been cosleeping compared with 21% of the random control infants.

Although the proportion of SIDS and control infants who slept in a room alone was similar (27% v 24%), relatively fewer SIDS infants compared with control infants (19% v 55%) slept in a cot next to their parents' bed. One quarter of the SIDS infants were swaddled before the last sleep, a bigger proportion than among

the controls (19% v 6%), who were usually swaddled in a single thin layer; 12 of these infants were found supine, four on their side, and three prone. More SIDS infants than control infants slept on a pillow; for half of these SIDS infants this included the entire body and various sleeping environments: a cot (n=6), the parents' bed (n=7), and cosleeping on a sofa (n=3). More of the SIDS infants than control infants (14% v 6%) were placed prone, but the difference was not significant (P=0.07). Nearly one third of the SIDS infants (29%) were found prone and over one quarter (28%) were described by the parents as in fair or poor health before the last sleep; both significant factors in the multivariable analysis. Significant background characteristics included maternal smoking during pregnancy, poor maternal education, preterm infants, and larger families. Several further socioeconomic factors or factors related to the significant findings were tested and despite being significant in the univariable analysis were not significant in the multivariable model. The variable unintentionally dropped, that had more than 5% of data missing and was added at the end of the modelling process, proved to be significant (P=0.006); 12 SIDS infants compared with two random control infants had been unintentionally dropped. Detailed information on the timing of these events was not recorded. None of these SIDS infants had any evidence of injury at postmortem examination.

Notable non-significant findings

Some findings that were statistically significant in the larger Confidential Enquiry into Stillbirths and Deaths in Infancy study 10 years ago²¹⁵⁻¹⁷ were notable by their lack of significance in the present study (table 3). A previous highly significant risk factor was SIDS infants with bedding over their head or face. This led to the "Feet to Foot" campaign in the UK in 1997, advising parents to place their infant's feet at the foot of the cot. Results (table 3) suggest that this advice has been followed and that the proportion of SIDS infants with their head covered has fallen significantly between studies, from 16% to 5% (P=0.01). Another reason for the difference in statistical findings may be the increasing practice of using infant sleeping bags,

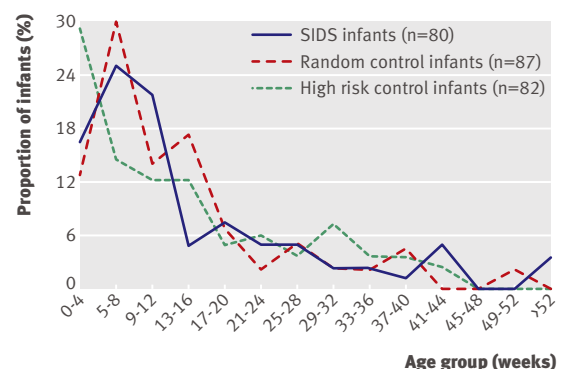


Fig 2 Age distribution of SIDS (sudden infant death syndrome) infants at time of death and of control infants at time of home visit and investigation of sleep scene

Table 2 | Multivariable logistic regression model comparing SIDS (sudden infant death syndrome) infants with random control infants. Values are numbers (percentages) unless stated otherwise

Risk factors for SIDS	SIDS infants	Random control infants	Multivariable odds ratio (95% CI)*	P value
Mother consumed >2 units† of alcohol in past 24 hours	19/77 (25)	2/87 (2)	41.62 (5.45 to 318.09)	0.0003
Infant shared parental bed or sofa for last sleep‡	43/79 (54)	18/87 (21)	21.77 (3.79 to 125.00)	0.001
Infant slept in room without parent for last sleep‡	21/79 (27)	21/87 (24)	21.34 (2.99 to 152.56)	0.002
Mother smoked during pregnancy	47/79 (59)	12/87 (14)	13.36 (3.07 to 58.83)	0.001
Infant swaddled for last sleep	19/78 (24)	5/87 (6)	31.06 (4.21 to 228.94)	0.001
Mother had no educational qualifications	28/80 (35)	12/87 (14)	15.55 (2.59 to 93.50)	0.003
Infant found prone for last sleep	23/79 (29)	9/86 (10)	6.61 (1.57 to 27.88)	0.010
Gestational age of infant ≤37 weeks	21/80 (26)	4/87 (5)	11.52 (1.64 to 80.82)	0.014
>3 live births (including this birth)	18/80 (23)	5/87 (6)	11.64 (1.57 to 86.05)	0.016
Infant (head or whole body) placed on pillow for last sleep	16/78 (21)	3/87 (3)	10.59 (1.43 to 78.39)	0.021
Infant's health in last 24 hours fair or poor	22/79 (28)	5/87 (6)	8.06 (1.11 to 58.42)	0.039
Maternal age at time of interview ≤21 years	24/80 (30)	15/87 (17)	—	0.053
Parental social class (on basis of occupation§) IV, V, or never employed	38/78 (49)	18/87 (21)	—	0.15
Housing not owned or mortgaged	60/76 (79)	37/87 (43)	—	0.17
Infant admitted to neonatal intensive care unit at birth	15/80 (19)	7/87 (8)	—	0.23
Parental use of narcotics in last 24 hours	11/78 (14)	5/87 (6)	—	0.28
Used infant sleeping bag for last sleep	2/77 (3)	11/86 (13)	—	0.42
Infant appeared ill in last 24 hours	26/80 (33)	6/87 (7)	—	0.64
Infant birth weight <2500 g	13/80 (16)	2/87 (2)	—	0.66
Used adult or infant duvet for last sleep	24/76 (32)	10/86 (12)	—	0.82
No waged income for household	28/77 (36)	15/87 (17)	—	0.89

Multivariable logistic regression model includes 74/80 SIDS cases (93%) and 86/87 controls (99%). Risk factors from variable maternal age at time of interview onwards were significant in univariable analysis but not significant in multivariable model; P value for these variables calculated by adding each variable to full model.

*Adjusted for infant's age (P=0.70) and daytime or night time sleep (p=0.74) as well as significant factors shown in multivariable model.

†Maximum recommended daily alcohol intake for women (United Kingdom): one unit is equivalent to one glass of beer (half pint), one small glass of wine, or one measure of spirits.

‡Multicategorical variable; infants sleeping in cot next to parental bed used as reference group.

§Choosing parental occupation closest to social class I (professional) or using maternal occupation if single mother. IV=partly skilled; V=unskilled.

which was significantly protective in the univariable analysis although not in the multivariable model (table 2). Other advice arising from the national study was to avoid placing infants on their side to sleep,³ to avoid excessive bedding and clothing,³ and to keep infants in a smoke-free zone.⁴ The proportion of infants placed on their side for the last sleep has more than halved between the two studies, among both the SIDS infants (P=0.0002) and the random control infants (P=0.0004). The proportion of infants exposed to tobacco smoke daily has fallen threefold among SIDS infants (P<0.001) and random control infants (P=0.0004). This may partly result from a real decline in maternal smoking over the past decade; the proportion of control mothers who smoked during pregnancy had decreased significantly, from 27% to 14% (P=0.007), although the decrease (66% to 60%) among the mothers of SIDS infants was smaller and not significant (P=0.29). The proportion of infants wrapped in 10 tog or more of clothes and bedclothes also fell significantly between studies for both the SIDS infants (P=0.007) and the control infants (P=0.009).

The prevalence of infants using dummies has decreased in the past decade and may be related to the increasing practice of breast feeding (table 3). The parents of eight SIDS infants and 16 control infants had observed the length of time the dummy stayed in the infant's mouth during the last or reference sleep; this was only a few minutes for four SIDS infants

and 12 control infants, with only one SIDS infant and one control infant retaining the dummy for more than one hour.

SIDS infants v high risk control infants *Significant multivariable findings*

The high risk control families were selected to reflect characteristics of the SIDS families for maternal smoking, social deprivation, maternal age, and larger family size (table 1). Given that the weighted factors were not tested for in the multivariable comparison between SIDS infants and high risk control infants (table 4) the results of the multivariable comparison between these infants were similar; important risk factors were maternal alcohol consumption, cosleeping, sleeping in prone position, being swaddled, preterm delivery, poor recent health, and use of a pillow.

Deaths while cosleeping

Variation in sleeping practice

Of the 80 SIDS infants, 79 died during sleep and one died in hospital after an operation. Table 5 lists the sleeping environment in which the SIDS infants were found and in which the controls awoke after the reference sleep. Most of the random and high risk control infants were in a cot by the parents' bed for the last sleep (51% and 55%). Most of the SIDS infants (54%) were found cosleeping with an adult, including 13 who coslept on a sofa, an uncommon environment among

Table 3 | Comparison of non-significant findings in SIDS (sudden infant death syndrome) infants and random control infants in south west England study and earlier national study. Values are numbers (percentages)

Factors	Current study (2003-6)		CESDI study (1993-6)	
	SIDS infants	Random control infants	SIDS infants	Random control infants
Infant found with bedding over head or face for last sleep	4/78 (5)	0/87 (0)	49/303 (16)	38/1289 (3)
Infant had feet at foot of cot for last sleep (cot sleepers only)	16/32 (50)	44/67 (66)	7/205 (3)	40/982 (4)
Infant placed on side for last sleep	14/77 (18)	9/87 (10)	129/317 (41)	361/1295 (28)
Infant exposed to tobacco smoke in postnatal period*	11/68 (16)	5/80 (6)	165/308 (54)	298/1288 (23)
Infant covered and clothed with ≥ 10 tog for last sleep	4/77 (5)	0/82 (0)	56/320 (18)	101/1299 (8)
Mother attempted to breast feed	55/79 (70)	69/87 (79)	141/323 (44)	774/1298 (60)
Infant used dummy for last sleep	11/70 (16)	18/87 (21)	124/313 (40)	664/1296 (51)

CESDI=Confidential Enquiry into Stillbirths and Deaths in Infancy.

*Parents' estimate of at least an hour a day inside or outside home.

the controls (one random control infant). Of the 13 SIDS infants who coslept on a sofa, six slept with the mother, six with the mother's partner, and one with an older sibling. For one SIDS infant this was the usual sleeping environment. The reasons given for cosleeping on a sofa for that particular night were that the infant would not settle ($n=1$), the infant needed feeding but the parent inadvertently fell asleep ($n=7$), the infant needed feeding but the parent decided to sleep ($n=1$), and the family was visiting or on holiday ($n=3$). In the case of the one random control infant who coslept on the sofa, the infant would not settle.

The median age of the SIDS infants varied with the last sleeping environment. SIDS infants who slept in a room alone or without parental supervision during daytime sleeps were typically around 6 months old (median 185 days, interquartile range 101-296 days), those who slept alone but shared the room with adults were just over 2 months old (64, 39-92 days), as were those infants who coslept on a sofa (69, 43-12 days), whereas infants who coslept in the parents' bed were the youngest (47, 18-72 days).

Cosleeping, alcohol, and drugs

None of the cosleeping parents of SIDS infants or control infants was taking prescribed drugs to help them sleep. Table 6 shows the proportion of parents who consumed more than two units of alcohol or took drugs before the infant's last sleep. Drugs included cannabis, heroin, methadone, cocaine, and amphetamines.

Overall, a greater proportion of parents of SIDS infants than those of control infants consumed alcohol and took drugs before the infant's last sleep, although this was significant only for mothers consuming alcohol. Combining the use of alcohol or drugs for either parent showed a significant difference between the parents of SIDS infants and those of random control infants, but this was not significant when comparison was made with parents of high risk control infants. The same pattern was observed when measuring regular alcohol and drug use, although the differences were not as striking (data not shown).

The contrast was more noticeable when the combined effect of cosleeping and alcohol or drug use was evaluated (table 7). No univariable risk was

associated with alcohol or drug use in the absence of cosleeping regardless of the control group used for comparison. The combination of alcohol or drug use before cosleeping was nine times more prevalent among the parents of SIDS infants than among those of the random control infants and six times more prevalent than among those of the high risk control infants. In nine of the families with SIDS infants and none of the control families the daily alcohol consumption was in excess of six units. Of the 24 SIDS infants who had coslept with an adult who had consumed more than two units of alcohol or taken drugs, the adult was the mother in 12 cases, the mother's partner in seven, and both parents in five. Drugs or alcohol were associated with seven of the 13 SIDS infants who coslept on a sofa. Interviews with control families were on weekdays; thus data are missing on drug and alcohol use on Fridays and Saturdays. When the analysis was limited to the deaths and reference sleeps that occurred in the 24 hours before the interviews on Mondays to Fridays the combined effect of cosleeping and alcohol or drugs among the SIDS infants was still highly significant compared with both the random control infants (odds ratio 10.73, 95% confidence interval 2.52 to 62.75, $P<0.001$) and the high risk control infants (7.74, 2.03 to 35.50, $P=0.0004$).

The combination of recent maternal alcohol consumption and cosleeping with an infant on a bed or sofa were the strongest predictors of SIDS in the multivariable model, regardless of which control was used as the comparison. Table 8 shows the results of the interaction between cosleeping and parents taking drugs or alcohol in the multivariable models, adjusting for both the weighting factors and risk factors previously reported to be a significant predictor of SIDS. Whether the families of random or high risk control infants were compared with those of the families of SIDS infants, no risk was associated with alcohol or drug use when the parents did not cosleep. Despite small numbers in the study the interaction between cosleeping and parental alcohol or drug use was significant when the families of SIDS infants were compared with those of the random control infants ($P=0.002$). This remained significant when the families of high risk control infants were used as the comparison ($P=0.02$). A decreased but significant risk was associated with cosleeping in the

Table 4 | Multivariable logistic regression model comparing SIDS (sudden infant death syndrome) infants with high risk control infants. Values are numbers (percentages) unless stated otherwise

Factors	SIDS infants	High risk control infants	Multivariable odds ratio (95% CI)*	P value
Mother consumed >2 units† of alcohol in last 24 hours	19/77 (25)	3/82 (4)	26.81 (4.36 to 164.99)	0.0004
Infant shared parental bed or sofa for last sleep‡	43/79 (54)	16/82 (20)	10.07 (2.80 to 36.24)	0.0004
Infant found prone for last sleep	23/79 (29)	7/81 (9)	11.47 (2.29 to 57.56)	0.003
Gestational age of infant ≤37 weeks	21/80 (26)	3/82 (4)	11.17 (2.22 to 56.35)	0.003
Infant swaddled for last sleep	19/78 (24)	3/82 (4)	10.67 (2.14 to 53.29)	0.004
Infant's health in last 24 hours fair or poor	22/79 (28)	7/82 (9)	6.53 (1.54 to 27.69)	0.01
Infant (head or whole body) placed on pillow for last sleep	16/78 (21)	3/82 (4)	8.47 (1.48 to 48.40)	0.02
Infant slept in room without parent for last sleep‡	21/79 (27)	17/82 (21)	—	0.08
Apgar score <8 at 1 minute	15/76 (20)	6/79 (8)	—	0.21
Infant admitted to neonatal intensive care unit at birth	15/80 (19)	2/82 (2)	—	0.31
Infant appeared ill in last 24 hours	26/80 (33)	6/82 (7)	—	0.41
Infant birth weight <2500 g	13/80 (16)	3/82 (4)	—	0.44

Multivariable logistic regression model includes 74/80 SIDS cases (93%) and 81/82 controls (99%). Factors from variable infant sleeping in room without parent onwards were significant in univariable analysis but not significant in multivariable model; P value of these variables calculated by adding each variable to full model.

*Adjusted for infant's age (P=0.77) and daytime or night time sleep (P=0.35), maternal smoking during pregnancy (P=0.18), maternal social class (P=0.04), young maternal age (P=0.86), and ≥3 live births (P=0.73) as well as other significant factors in multivariable model.

†Maximum recommended daily alcohol intake for women (United Kingdom): one unit is equivalent to one glass of beer (half pint), one small glass of wine, or one measure of spirits.

‡Multicategorical variable; infants who slept in a cot next to parental bed were reference group.

absence of alcohol or drug use, although this included infants who coslept on a sofa. The proportion of SIDS infants found cosleeping in a bed with parents who had drunk two units or less of alcohol and taken no drugs was no different from that of the random control infants (18% *v* 16%). If parents who regularly smoked were further excluded, then five of the SIDS infants (6%) were found in this less risky cosleeping environment compared with nine of the random control infants (10%).

DISCUSSION

Many of the deaths in this case-control study of SIDS in the south west of England occurred while the infants coslept in a hazardous environment. The major influences on risk of SIDS, regardless of markers for socioeconomic deprivation, are amenable to change and specific advice needs to be given, particularly on the use of alcohol or drugs if cosleeping with an infant and the risk of cosleeping on a sofa.

Strengths and limitations of the study

The noticeable reduction in SIDS rates and the consequent relatively small number of deaths from SIDS in this study limits the interpretation of complex multivariable relations between contributory factors and may underestimate the significance of some factors. Those found to be significant in this study are, however, highly likely to be real differences as are any significant interactions between them. Given the reduction in SIDS rates, it is unlikely that larger population based case-control studies will be possible in the future.

The difficulty in recruiting control families may also limit the interpretation of the results; weighting the random process by socioeconomic status may offset some of the selection bias but does not rule out the possibility that our random control families were different from the population by measures other than occupational

status. Our second control group, that of high risk families, was therefore important as not only were their characteristics more similar to those of the families with SIDS infants but they also had more in common with deprived families—a group potentially under-represented in control groups in previous studies.

Another limitation is that we did not interview the control families at weekends, when alcohol and drug use may be more common. However, an analysis restricted to deaths and reference sleeps that occurred only during the week suggests the combined effect of cosleeping and alcohol or drug use was still highly significant.

Despite these limitations, that the differences for almost all factors investigated were similar in magnitude between the SIDS infants and the high risk control infants to those observed between SIDS infants and the randomly selected control infants is important. Despite their similar socioeconomic backgrounds, the high risk control infants were as different from the SIDS infants in many important risk factors as were the random control infants, confirming that the risk factors for SIDS are not merely surrogate markers for aspects of social deprivation.

The more recent reduction in SIDS rates is difficult to estimate from national figures because of a recent trend among coroners and pathologists in the UK to label such deaths as “unascertained” if overlaying or intentional injury is suspected but not proved.¹⁸ This diagnostic shift has been recognised by the Office for National Statistics.¹⁹ The estimates from our two studies are, however, population based with virtual complete ascertainment using a consistent diagnosis throughout the region. The study estimates confirm the continued and significant (P<0.001) fall in incidence of SIDS deaths from 1 week to 1 year old in the decade that followed the “Back to Sleep” campaign; from 0.77 per 1000 live births in 1993-6 to 0.45 per 1000 live births in 2003-6.

Table 5 | Sleeping environment in which SIDS (sudden infant death syndrome) infants and control infants were found for last sleep. Values are numbers (percentages)

Sleeping environment for last sleep	SIDS infants	Random control infants	High risk control infants
In cot*:			
Parent in room	12/79 (15)	44/87 (51)	45/82 (55)
Parent not in room	18/79 (23)	18/87 (21)	16/82 (20)
In parental bed:			
Cosleeping with adult	30/79 (38)	17/87 (20)	16/82 (20)
Sleeping alone†	2/79 (3)	3/87 (3)	1/82 (1)
On sofa:			
Cosleeping with adult	13/79 (16)	1/87 (1)	0/82 (0)
Sleeping alone, with parent in room	2/79 (3)	0/87 (0)	0/82 (0)
Other:			
Pushchair, baby chair, or bouncy chair‡	2/79 (3)	4/87 (5)	3/82 (4)
Floor, with parent in room	0/79 (0)	0/87 (0)	1/82 (1)

*Including cradle, crib, or Moses basket.

†For one SIDS infant and one random control infant parent was present in room.

‡For three random control infants and three high risk control infants parent was present in room.

Factors contributing to reduction in SIDS rate

The proportion of both SIDS and control infants put or found in the prone position were no different from those in our earlier Confidential Enquiry into Stillbirths and Deaths in Infancy study, suggesting that the reduction in SIDS rates may be caused by something else. One possible contributing factor may be the reduction in prevalence of infants sleeping on their side, which has more than halved among both SIDS infants and control infants compared with such infants in our earlier study. Also, notably fewer infants are now exposed to environmental tobacco smoke, at least a threefold reduction among both SIDS infants and randomly chosen controls, although maternal smoking during pregnancy remains an important risk factor.

Factors that have changed and may have contributed to the reduction in SIDS rates over the past decade include a reduction in the thermal insulation of bedding, together with widespread uptake of the “feet to foot” message, and a noticeable increase in the use of infant sleeping bags. Both are designed to prevent the head from being covered, a factor highly significant in previous studies¹⁷ but which was significantly less common among SIDS infants in the present study

Table 6 | Alcohol or drug consumption by parents of SIDS (sudden infant death syndrome) infants or control infants before last sleep. Values are numbers (percentages)

Variable	SIDS infants	Random control infants	High risk control infants
Alcohol >2 units‡:			
Mother	19/77 (25)	2/87 (2)***	3/82 (4)***
Partner	18/78 (23)	15/87 (17)	17/82 (21)
Drugs†:			
Mother	5/77 (6)	2/87 (2)	2/82 (2)
Partner	7/76 (9)	4/86 (5)	4/82 (5)
Alcohol >2 units† or drugs‡			
Either parent	30/79 (38)	20/87 (23)*	22/82 (27)

*P<0.05; **P<0.01; ***P<0.001.

†Maximum recommended daily alcohol intake for women (United Kingdom): one unit is equivalent to one glass of beer (half pint), one small glass of wine, or one measure of spirits

‡Methadone, cannabis, or amphetamines.

compared with the Confidential Enquiry into Stillbirths and Deaths in Infancy study (5% v 16%, P=0.02). The thermal insulation of bedding plus clothing used in both SIDS infants and control infants has fallen progressively, from 10 tog and 8 tog, respectively, in the Avon studies in the 1980s²⁰ to 5 tog and 4 tog in the Confidential Enquiry into Stillbirths and Deaths in Infancy study,³ and to 3 tog and 2.4 tog in the present study.

New factors emerging

The risk associated with placing an infant's head or whole body on a pillow has become more pronounced than in our previous study and was prevalent among the various sleeping environments in which the SIDS infants were found. Swaddling infants may be a potential new risk factor; a recent review suggests that this may be a risk factor only in combination with the prone sleeping position,²¹ although most swaddled SIDS infants in this study were found supine.

The reduction in the peak age of deaths in the present study is notable, with the SIDS rate among older infants having fallen more than among the younger infants. This may be important in understanding the increased proportion of deaths while bed sharing, as such deaths have previously been reported to be more common among younger SIDS infants.^{8,11}

Dummy use and SIDS

The unexpected decline in the prevalence of dummy use among both SIDS infants and control infants may be related to the increase in breast feeding among mothers of both SIDS infants (70% attempted in this study compared with 44% in the Confidential Enquiry into Stillbirths and Deaths in Infancy study) and random control infants (79% and 60%, respectively). The data on the length of time the dummy remained in the infant's mouth, although limited, suggest no difference between the groups and that the dummy fell out of the infant's mouth soon after sleep, casting doubt on the idea that the act of sucking affords protection. The fall in SIDS rate despite a fall in prevalence of dummy use does not support the hypothesis that dummies are directly involved in a potentially protective mechanism against SIDS.²²

Measuring cosleeping and alcohol or drug use in previous studies

Several epidemiological studies on SIDS have found either no evidence or weak evidence of a risk from parents habitually consuming alcohol.^{4,23-27} Alcohol consumption by parents immediately before the death of their infant or the reference sleep has been investigated in fewer studies and either has not been measured in conjunction with cosleeping²⁸ or has pointed towards an association, although the evidence remained anecdotal because of a lack of data on controls.²⁹⁻³¹ Our controlled observations 10 years ago¹¹ suggested a potential link between cosleeping and recent alcohol consumption, although we did not

Table 7 | Association between recent alcohol or drug use by parents and cosleeping with SIDS (sudden infant death syndrome) infants or random and high risk control infants. Values are numbers (percentages) unless stated otherwise

Parental behaviour		SIDS infants	Control infants	Univariable odds ratio (95% CI)	P value
Recent alcohol or drug use*	Coslept with infant†				
Random control infants					
No	No	29/78 (37)	52/87 (60)	1.00 (reference)	
Yes	No	6/78 (8)	17/87 (20)	0.63 (0.18 to 1.93)	0.54
No	Yes	19/78 (24)	15/87 (17)	2.27 (0.93 to 5.57)	0.07
Yes	Yes	24/78 (31)	3/87 (3)	14.34 (3.78 to 78.76)	<0.001
High risk control infants					
No	No	29/78 (37)	50/82 (61)	1.00 (reference)	
Yes	No	6/78 (8)	16/82 (20)	0.65 (0.19 to 2.00)	0.57
No	Yes	19/78 (24)	12/82 (15)	2.73 (1.07 to 7.03)	0.03
Yes	Yes	24/78 (31)	4/82 (5)	10.34 (3.06 to 44.10)	<0.001

*Consumed more than two units of alcohol (one unit is equivalent to one glass of beer (half pint), one small glass of wine, or one measure of spirits) or took methadone, cannabis, or amphetamines before last sleep.
†In parental bed or on sofa.

collect data on recent drug use. Published data on the use of drugs and the risk of SIDS are even more sparse, and more difficult to collect given the often illegal nature of their use. Studies have tended to concentrate on the effects in utero³²⁻³⁴ and the potential risk associated with habitual use³⁵ rather than investigating any interaction with cosleeping.

Cosleeping and alcohol or drug use

The findings suggest that much of the risk associated with cosleeping may be explained by the circumstances in which the SIDS infants were found. A significant risk was associated with cosleeping on a sofa or on any surface with a parent who had consumed alcohol or drugs. In the absence of any evidence that the parent had laid on the infant, from investigation of the scene and circumstances or from postmortem examination, it is a simplistic and unjustified assumption that all unexpected deaths in potentially risky cosleeping environments are caused by overlaying or entrapment. Indeed in this investigation of all sudden unexpected deaths in infants the multiprofessional review process attributed only three deaths to unintentional asphyxiation. These deaths were thus not labelled as SIDS and

are not included in the present analysis. It would be wrong to apportion blame on an individual basis without sufficient evidence, but this does not mean we can ignore the patterns we observe at a population level. Almost regardless of the pathophysiological processes leading to infant deaths in risky cosleeping environments we should remind parents that such cosleeping practices are risky. The increased risk of unintentional suffocation in such circumstances needs to be reinforced.

Despite the small numbers in this study the interaction between cosleeping and recent use of alcohol or drugs by the parents remained significant in the multivariable analysis. This finding was significant regardless of which control group was used for comparison, suggesting that we are not merely measuring aspects of deprivation but identifying specific circumstances that put infants at risk.

The strong association between alcohol consumption, use of drugs, and smoking may explain in part the interaction found previously between cosleeping and smoking.^{8 11 24} Our findings may also partially explain the difference in SIDS rates between cultures where cosleeping is the usual practice. In certain cultures bed sharing is common and the prevalence of SIDS is high. These include the African black populations in the United States and Maori and Aboriginal populations. Intriguingly, the SIDS rates are low in other cultures where bed sharing is common, including Japan and Hong Kong, Bangladeshi and other Asian communities in the UK, and Pacific Islander communities in New Zealand. It is not bed sharing that distinguishes these cultures but other mediating factors such as smoking and use of alcohol and drugs, which in conjunction with cosleeping may put infants at risk.³⁶

Cosleeping on sofas

Using our longitudinal data from Avon over the past 20 years we have already shown an increase in infant deaths while cosleeping on a sofa. The proportion of cosleeping SIDS infants found on a sofa has increased significantly ($P=0.0003$) between our case-control studies, from 6% in 1993-6 to 16% in this study. It is the only infant sleeping environment in which the SIDS

Table 8 | Multivariable logistic regression model to test for interaction between cosleeping and recent alcohol and drug use by parents of SIDS (sudden infant death syndrome) infants and random or high risk control infants

Factors	Model 1: SIDS infants v random control infants*		Model 2: SIDS infants v high risk control infants†	
	Odds ratio (95% CI)	P value	Odds ratio (95% CI)	P value
Infant coslept in parental bed or on sofa for last sleep	5.41 (1.12 to 26.17)	0.04	5.23 (1.37 to 19.91)	0.02
Parental use of alcohol (>2 units‡) or drugs before last sleep§	0.52 (0.10 to 2.72)	0.44	0.69 (0.16 to 3.00)	0.62
Interaction between cosleeping and alcohol or drug use	53.26 (4.07 to 696.96)	0.002	11.76 (1.40 to 99.83)	0.02

Multivariable logistic regression models includes 74/80 SIDS cases (93%), 86/87 random controls (99%), and 81/82 high risk controls (99%).

*Multivariable model adjusted for weighting factors infant age and daytime or night time sleep along with significant risk factors; maternal smoking during pregnancy, maternal education, infant found sleeping prone, infant swaddled, gestational age, not sharing room, and fair or poor health in last 24 hours.

†Multivariable model adjusted for weighting factors infant age, daytime or night time sleep, maternal smoking during pregnancy, number of live births, young maternal age, and poor socioeconomic status along with infant found sleeping prone, infant swaddled, infant placed on pillow, gestational age, not sharing room, and fair or poor health in last 24 hours.

‡One unit is equivalent to one glass of beer (half pint), one small glass of wine, or one measure of spirits.

§Methadone, cannabis, or amphetamines

WHAT IS ALREADY KNOWN ON THIS TOPIC

Since we carried out an English study on sudden infant death syndrome (SIDS) 10 years ago the SIDS rate has continued to decrease

It is unclear which risk reduction messages have contributed towards this continued fall in rates

A higher proportion of the residual SIDS deaths now occur among more deprived families and those who cosleep with their infant

WHAT THIS STUDY ADDS

Many of the deaths while cosleeping occurred in potentially hazardous environments, including a sofa or shared surface with an adult who had recently consumed alcohol or narcotics

The importance of covering an infant's head during sleep, postnatal exposure to tobacco, use of a dummy, and sleeping in the side position has diminished, although being found prone was still a risk factor

The significance of swaddling infants and the use of pillows has increased

Advising parents to avoid risky cosleeping environments might reduce the SIDS rate even further

rate has increased in recent years, and it equates to an increase from 24 to 42 deaths a year in England and Wales during a period when the SIDS rate has halved; about one sixth of SIDS infants are now found cosleeping on a sofa. Similar proportions have been reported in Scotland⁵ and Northern Ireland³⁷ but were not found in a recent German study,³⁸ which may suggest this practice is culture specific. Cosleeping on a sofa was an uncommon practice among the control families, and it was not a regular practice even among the parents of 13 SIDS infants in this study. Alcohol or drugs were a feature in half of these deaths, and in seven cases the parents wanted to feed their infant and inadvertently fell asleep. This raises two important points. Firstly, it is not enough to advise against cosleeping on a sofa; health professionals must advise parents to avoid putting themselves in the position where this could happen. Secondly, any advice to discourage bed sharing may carry with it the danger of tired parents feeding their baby on a sofa, which carries a much greater risk than cosleeping in the parents' bed. Anecdotally, two of the families of SIDS infants who had coslept on a sofa informed us that they had been advised against bringing the baby into bed but had not realised the risks from falling asleep on the sofa.

Conclusions

Although socioeconomic markers were more prevalent among the families of SIDS infants, the major influences on risk were from factors amenable to change within the infant's sleeping environment. Some of the risk reduction messages seem to be getting across and may have contributed to the continued fall in the SIDS rate. Identifying emerging dangers and re-emphasising ones already observed within the infant sleeping environment may further reduce the number of deaths from SIDS. This is clearly illustrated in the current polarised debate surrounding cosleeping.

The safest place for an infant to sleep is in a cot beside the parents' bed. Based on evidence from research into SIDS it is questionable whether advice to avoid bed sharing is generalisable and whether such a simplistic approach would do no harm. Parents of young infants need to feed them during the night, sometimes several times, and if we demonise the parents' bed we may be in danger of the sofa being chosen. A better approach may be to warn parents of the specific circumstances that put infants at risk. Parents need to be advised never to put themselves in a situation where they might fall asleep with a young infant on a sofa. Parents also need to be reminded that they should never cosleep with an infant in any environment if they have consumed alcohol or drugs.

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