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CHILD HEALTH

Infant sleeping position and the sudden infant death syndrome: systematic review of observational studies and historical review of recommendations from 1940 to 2002

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Background Before the early 1990s, parents were advised to place infants to sleep on their front contrary to evidence from clinical research.

Methods We systematically reviewed associations between infant sleeping positions and sudden infant death syndrome (SIDS), explored sources of heterogeneity, and compared findings with published recommendations.

Results By 1970, there was a statistically significantly increased risk of SIDS for front sleeping compared with back (pooled odds ratio (OR) 2.93; 95% confidence interval (CI) 1.15, 7.47), and by 1986, for front compared with other positions (five studies, pooled OR 3.00; 1.69–5.31). The OR for front vs the back position was reduced as the prevalence of the front position in controls increased. The pooled OR for studies conducted before advice changed to avoid front sleeping was 2.95 (95% CI 1.69–5.15), and after was 6.91 (4.63–10.32). Sleeping on the front was recommended in books between 1943 and 1988 based on extrapolation from untested theory

Conclusions Advice to put infants to sleep on the front for nearly a half century was contrary to evidence available from 1970 that this was likely to be harmful. Systematic review of preventable risk factors for SIDS from 1970 would have led to earlier recognition of the risks of sleeping on the front and might have prevented over 10 000 infant deaths in the UK and at least 50 000 in Europe, the USA, and Australasia. Attenuation of the observed harm with increased adoption of the front position probably reflects a 'healthy adopter' phenomenon in that families at low risk of SIDS were more likely to adhere to prevailing health advice. This phenomenon is likely to be a general problem in the use of observational studies for assessing the safety of health promotion.

Keywords Sudden infant death, review, meta-analysis

Sudden unexpected unexplained infant death, now known as sudden infant death syndrome (SIDS), was recognized as a major cause of infant death in the UK and USA throughout the 20th century. At the start of the 20th century, such deaths were attributed to overlying, particularly by drunken mothers.[1] By the 1940s, as more deaths were investigated by autopsy, pathologists realized that few deaths were due to maternal overlying, and alternative mechanisms for 'accidental mechanical suffocation' were sought. In 1944, Abramson, a pathologist in New York State, noted that two-thirds of infants dying from mechanical suffocation were found face down, contrary to the usual sleeping position for infants at the time.[2] His observations, which were corroborated by reports in the UK

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and Australia[3,4] led to a health promotion campaign that recommended avoidance of the front position.[5]

The campaign was short-lived. In 1945, a paediatrician, Woolley, rejected Abramson's hypothesis of suffocation on the front based on experiments in which he had covered babies' faces with layers of blankets.[6] He reported that the oxygen content of the air breathed by the babies only fell when they were covered with a rubber sheet and that babies moved if breathing was obstructed. He also criticized the explanation of suffocation because it 'instilled guilt and self-incrimination in parents'.

Emergence of alternative explanations for death, such as unrecognized infection[4,7,8] inhalation of vomit[9] and hypersensitivity reaction to inhaled milk,[10] further strengthened the argument against the suffocation hypothesis and highlighted the need for data on risk factors. The first published case–control study was started in 1956 in the USA,[11] and in 1958, a similar study in the UK was the first to measure infant sleeping position in SIDS victims and live control babies.[12] At around the same time, it became increasingly common to advocate sleeping on the front. We now know that front sleeping is a major cause of SIDS. We wanted to know whether systematic review of the evidence could have reversed this harmful advice sooner or whether variation in the association between sleeping on the front and SIDS was consistent with recommendations at the time. We did a systematic review and meta-analysis of the effect of front and side sleeping on the risk of SIDS, and an historical review of recommendations on infant sleeping position in books and pamphlets on infant care available in the UK between 1940 and 2002. We focussed on how the strength of the evidence for a harmful effect of front sleeping changed before and after advice changed in favour of avoidance of the front position. We hypothesized that the effect of the front position on SIDS might differ depending on whether health advice favoured front or not as families that adopt health advice are likely to be at lower risk of SIDS.

Methods

Historical review

We reviewed recommendations on infant sleeping position in books or pamphlets available in the UK from 1940 to 2002. We chose 1940 to include a period before the front position was widely advocated. We searched the Modern Medicine Collection at the Wellcome Trust library, and, because of a lack of more recent texts, the British Medical Association library from 1965 to 2002. We included any book or pamphlet that referred to the care of normal term infants aged <6 months, and mentioned infant sleeping position. Searches used the library indexing system for books on infant care and we also searched electronically using terms for paediatric, parent, and baby (details of search strategy available from authors).

One reviewer (S.S. or M.H.), assessed whether texts met the inclusion criteria and prepared a hard copy file with the extract and book title but not the date of publication. A second reviewer (R.G.) categorized the recommendation as favouring front, back, side, or non-front position(s), or neutral if all or none were implicitly or explicitly favoured. A second reviewer (S.S.), independently categorized one-third of the texts and there was complete agreement with the first reviewer.

Systematic review

We included any case–control or cohort study that compared the risk of SIDS in infants sleeping on their front, side, or back. Studies had to be based on SIDS infants and live healthy control infants from the same community. We searched for any comparative study of infant sleeping position and SIDS in MEDLINE (1966–2002) and EMBASE (1980–2002), using a detailed search strategy (available from the authors), and reference lists of review articles, a PhD thesis on the history of SIDS,[13] and included studies. Abstracts were scanned by one reviewer (S.S., M.H., or R.G.), and full texts of potentially eligible studies retrieved. R.G. and S.P. jointly extracted data from included studies.

Data quality

We used data on the position in which the infant was placed to sleep before death or interview, or if lacking, data on usual position, or position found. If usual position was measured at multiple ages, we used results closest to 3 months of age. We recorded the method of selection of cases and controls, matching criteria, if any, and whether data collection methods differed in cases and controls.

Analysis

Our primary aim was to compare the risk of SIDS in infants sleeping front and back. As some studies did not separately report side and back positions, we also compared front with non-front positions. However, grouping side with back will attenuate the observed risk associated with the front position if the side position is also harmful. We therefore calculated odds ratios (ORs) for SIDS associated with sleeping front vs back, front vs non-front, and side vs back.

To avoid confounding, we used the unadjusted matched OR if reported. Otherwise we calculated the unmatched OR.[14] Because studies differed in their design, populations, and methods, we used a random effects model in which it is assumed that the observed ORs are sampled from a common distribution around a mean effect with variance measured by the heterogeneity parameter. We estimated 95% confidence intervals (CIs) and considered a P-value <0.05 as statistically significant. Heterogeneity in the OR for SIDS was assessed by the chi-squared test (Q-test) and quantified using I^2 which reflects the proportion of variation that is not due to sampling error.[15] The possibility of publication bias was evaluated using funnel plots and the Egger and Begg tests.[16,17]

We determined the year at which there was a statistically significant association between front or side sleeping positions and SIDS by using a cumulative meta-analysis based on year of publication as described by Lau.[18] The overall heterogeneity was used in the calculation of the CIs for the cumulative OR at every step using a random effects model. We applied recursive cumulative meta-analysis to examine the direction and magnitude of the relative changes in the cumulative evidence as a function of the cumulative sample size.[19,20] At the end of every information period j, the ratio (cumulative OR_j)/(cumulative OR_{j+1}) was assessed and compared with unity. If larger than one, this was interpreted as a 'move' of the evidence towards defining the front position as more harmful than in the previous information period.

To explore potential sources of heterogeneity we initially used conventional meta-regression to determine an association with variables previously suggested.[21] In a univariate model, we first determined the effect of the position recorded in cases (before death, usual, or after death), year of publication, recruitment year (measured as the mid-point between start and end of recruitment), matching criteria for controls and cases, and country and continent of study. The combined effect on heterogeneity of the variables found to be significant in the univariate analysis was estimated in a multivariate meta-regression model. We extended the meta-regression analyses to examine the hypothesis that the prevalence of front sleeping in control infants is associated with heterogeneity. This is because parents who put their babies to sleep in the front position when advised not to, might have a different risk of SIDS than parents who do so when front sleeping is the norm (similarly for the side position).

The OR for front vs any other position can be written as using $\text{logOR} = \text{logit}P(\text{front}|\text{case}) - \text{logit}P(\text{front}|\text{control})$ and the prevalence of front sleeping is estimated in the controls as $P(\text{front}) = P(\text{front}|\text{control})$. Consequently, regression of logOR to $P(\text{front})$ will be biased by regression to the mean.

To overcome this we fitted a hierarchical model similar to that described by Thompson *et al*. to model background risk in randomized controlled trials.[22,23] As the studies were case-control rather than trials, we made some modifications to the methods (see Appendix).

We retained in the model any factors that were statistically significantly associated with heterogeneity in the conventional meta-regression, and assessed the extent to which the factors included in the model explained the variation between studies by measuring the change in the heterogeneity parameter. If factors included in the model explained heterogeneity, the heterogeneity parameter (variance in the random effects) would be expected to get smaller. This model was fitted using Markov chain Monte Carlo methods within a Bayesian framework. The analysis was conducted using Intercooled Stata 8.2 (Stata Corp., College Station, TX), R 1.9.1 (R Foundation for statistical computing, Vienna) and Winbugs 1.4.1.

Results

Historical review

Table 1 summarizes the recommendations made in 83 texts that met the inclusion criteria (details available from the authors). From 1940 to the mid-1950s, texts favoured the back or side positions and only one, in 1943, recommended the front position. From 1954 until 1988, a substantial proportion of texts consistently favoured front sleeping, although many also favoured the side and back. The sudden shift in favour of front sleeping is best illustrated by 'Baby and Child Care' by Dr Benjamin Spock who recommended the back position in his 1955 edition, and the front position in 1956.[24] In his 1958 edition, he argued 'If he vomits, he's more likely to choke on the vomitus. Also, he tends to keep his head turned to the same side—usually toward the centre of the room. This may flatten the side of his head.' Many authors repeated these arguments. Others argued that front sleeping reduced wind,[25,26] coughing due to mucus,[27] and made respiration easier.[26] Suffocation was considered to be possible only if the baby was very weak.[26] These views were not universal. In editions of his textbook in 1945, 1950, and 1959, Nelson stated that 'position during sleep

Table 1 Recommended infant sleeping position in books on infant care

Year	No. texts	Front	Side	Back	Non-front	Neutral
1940	1		●			
	0					
1942	1		●			
	2	●	●	●●		
1944	1		●			
	1					●
1946	5		●●	●●**		
	1		●	●		
1948	1			●		
	2		●●	●		
1950	1					●
	0					
1952	0					
	2		●●	●●		
1954	2					●●
	3		●	●*		
1956	2	*				●
	2			●		●
1958	2	*	●			
	2		●			●
1960	2	●	●			●
	0					
1962	1			●		
	1	●	●			
1964	2					●●
	0					
1966	1		●	●		
	2	●	●	●		
1968	3	●		●		●
	1	*				
1970	0					
	0					
1972	2					●●
	0					
1974	1	●		●		
	2	●		●		●
1976	0					
	1		●			
1978	0					
	2	●*	●			
1980	2	●	●	●		
	1	●				
1982	0					
	2		●	●		
1984	1	●	●			
	1		●			
1986	1	●	●			
	0					
1988	1	●	●			
	0					
1990	1			●		
	2		●●			
1992	3		●		●●	
	1				●	
1994	2				●●	
	2			●	●	
1996	2			●	●	
	1				●	
1998	3			●●●		
	3			●●●		
2000	0					
	1				●	
2002	1			●		

* Books written by Dr Benjamin Spock.

is relatively unimportant, but should be changed often to prevent moulding of the cranium'.[28–30] Others were less equivocal. One author recommended in 1953, 'Sleeping on his abdomen never should be permitted because of the danger of suffocating'.[31] In 1966, another warned 'Very small babies should never be left alone lying on their tummies. This is an American fashion to strengthen the back, but we think the dangers of suffocation are not sufficiently remote to justify it.'[32]

No texts favoured the front position after 1988. From the mid-1950s to 1990, many texts continued to recommend the side position, but few advocated sleeping on the back. In the early 1990s, most texts recommended the side position or simply advised against front sleeping, but apart from one text in 1990, the back position was not consistently advocated until 1995.

Systematic review

Of the 2897 abstracts scanned, and the 206 full text articles retrieved, 40 studies met the inclusion criteria (Figure 1 and Table 2). Four further studies were excluded (Figure 1). No randomized controlled trials were found. All 40 included studies provided data on front vs non-front positions, but only 24 studies separately recorded back and side positions. Of the 40 studies, 23 (and 15/24 reporting side and back positions) included some degree of matching of controls with cases. Of these, unadjusted matched ORs were available for 9/23 studies (and for 7/15 reporting side and back positions).[33,33–44] For one study, we derived pooled ORs from data reported for separate ethnic groups.[37] All studies were case–control except for one cohort study reported in two stages. This resulted in data for 2 years of the study (15 SIDS victims) being included twice in the cumulative meta-analyses.[45,46] Repeated use of the same data was avoided for all the other studies except for Mitchell 2 1999 (details in Table 2). No substantial evidence was found for publication bias for any of the sleeping position comparisons either by examining the funnel plots or applying the Egger or Begg tests (lowest *P*-value = 0.103).

There was a statistically significantly higher risk of death associated with the front position whether compared with the back (Figure 2a) or non-front positions (Figure 2c). There was a weak association between the side position and the risk of SIDS, which was marginally worse than back (Figure 2e).

The cumulative meta-analyses showed that the association between death and the front position compared with back had become statistically significant by 1970, after the first two case–control studies (cumulated OR 2.93; 95%CI 1.15–7.47; Figure 2b). When front was compared with non-front, the association was not statistically significant until 1986, after inclusion of five studies (cumulated OR 3.00; 1.69–5.31;

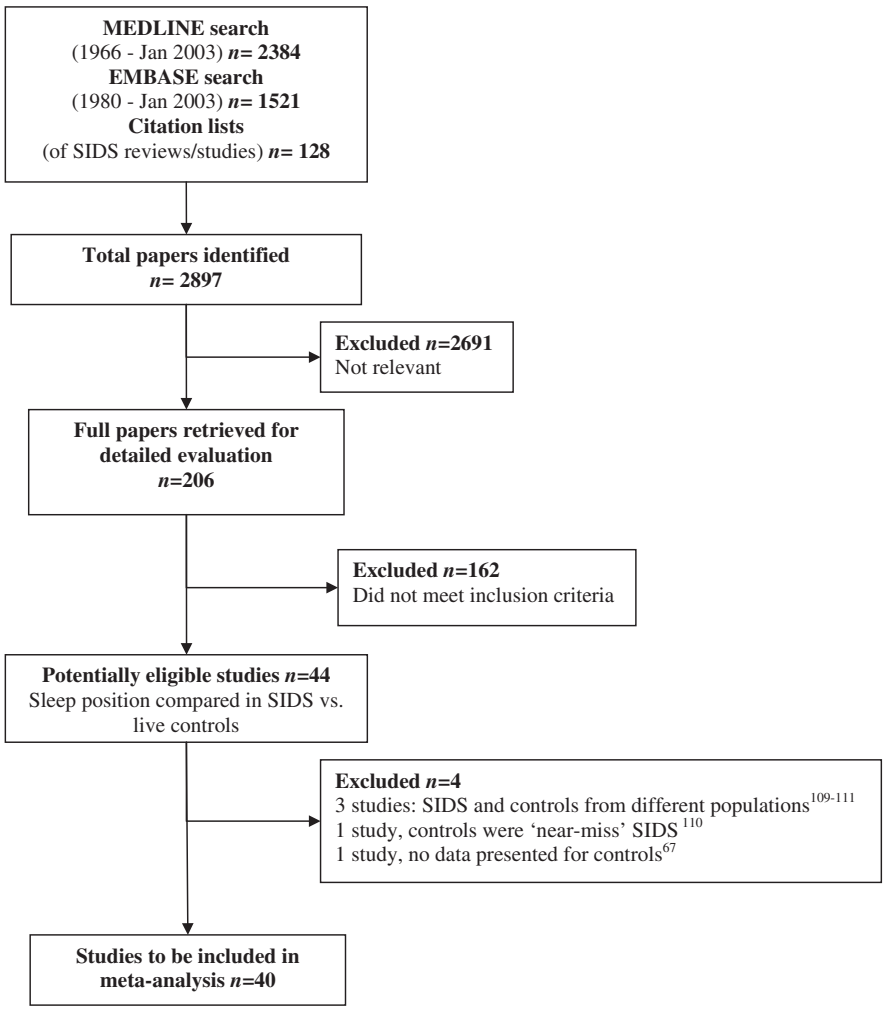

Figure 1 Flow diagram to show results of searches for the systematic review of comparative studies of infant sleeping position and SIDS

Table 2 Characteristics of included studies

Author, year published	Study period, selection of cases and controls, and method of data collection	Position recorded[d]	Back (%)	Front (%)	Total studied
Carpenter 1965[12a]	1958–1961. SIDS: Referred to coroner in 12 London boroughs (aged 2 weeks – 2 years, 6 of 100 SIDS victims were aged >12 months). Position found recorded by coroner.	3	20	25	107
	Controls: matched for age, sex and community from register of Medical Officer of Health. Sleeping position recorded by health visitors.	2	45	11	183
Froggatt 1970[57]	1965–1967. SIDS: Consecutive cases in northern Ireland.	2	11	7	139
	Controls: matched for age, sex, and administrative area. Data collection by home interviews in both cases and controls	2	35	4	143
Beal 1 1986[62]	1970–1984. SIDS: South Australia interviewed within weeks of death by Beal.	2	8	85	133
	Controls: postal survey of 200 consecutive birth registrations in August 1984.	2	23	39	152
Tonkin 1 1986[61]	1972–1982. SIDS: position found routinely recorded in Auckland, New Zealand in 1972, 1973, and 1982.	3	9	56	91
	Controls: Plunket nurses (health visitors) in Auckland noted sleeping position of 50 babies most recently seen (10 nurses in 1972, 15 nurses in 1973). In 1982 all nurses noted sleeping position of 2 week old babies during a 3 month period.	5	4	29	1982
Cameron 1986[59]	1980–1982. SIDS: within the Melbourne statistical division, Australia.	2		69	208
	Controls: matched by age and same hospital of birth. Data collection by home interviews in both groups.	2		41	393
Senecal 1987[93]	1984–1985. SIDS: in the Departement d'Ille et Vilaine, France. No details on how data were collected.	1		85	20
	Controls: Infants born in 7 maternity hospitals in Brittany attending routine post-natal surveillance. Questionnaire completed by doctor at consultation.	2		29	318
Nicholl 1988[33]	1976–1979. SIDS: UK multicentre study. SIDS resident within local areas.	2		42	265
	Controls: matched for age and area. Data collection by home interviews in both groups.	2		25	273
McGlashan 1989[58]	1980–1986. SIDS: notified by coroners in Tasmania.	2	5	59	164
	Controls: matched for age, sex, and hospital of birth. Data collection at home interviews in both groups.	2	10	43	329
Beal 2 1991[94]	1985–1989. SIDS: in South Australia interviewed within weeks of death by Beal.	2		80	100
	Controls: postal survey of 200 consecutive birth registrations in August in 1988.	2		29	182
Jonge 1989[63]	1980–1986. SIDS: deaths in The Netherlands.	2		86	142
	Data collected by home interview Controls: Infants at 17 well-baby clinics. Parents interviewed about sleeping position at 2–4 months and 5–7 months.	2		62	320
Tonkin 2 1989[64,65]	1981–1985. SIDS: deaths in Auckland, New Zealand.	1	13	54	126
	Data collected at interview. Controls: surveyed by plunket nurses in Auckland in 1983 aged 1–4 months. Results used for usual position at 3 months.	2	8	51	1138
Lee 1988[95]	1986–1987. SIDS: prospective surveillance of all SIDS deaths in Hong Kong. Data collected at home interview	2	56	44	16
	Controls: age and sex matched, one from hospital and one from community. No details given on data collection.	2	94	6	32
Fleming 1 1990[34]	1987–1989. SIDS: all SIDS in Avon, UK, interviewed at home within days of death.	2	1	93	67
	Controls: matched by age and area based on same health visitor list as SIDS victim. Data collection at home interview for both groups.	2	18	58	131
Bouvier-Colle 1990[96]	Study period not stated. SIDS: study in France, reported only in conference proceedings.	3		88	782
	Controls: no details available on selection or data collection.	2		34	211
Hoffman 1992[84]	1978–1979. SIDS: All SIDS in six geographically defined areas in the USA.	2		81	757
	Controls: matched for age, and study centre (second controls matched for ethnic group and birth weight not used in this review). Both SIDS and control parents were interviewed about usual sleeping position in the 2 weeks before death or interview.	2		72	757
Engelberts 1991[83]	1985–1987. SIDS: all deaths in The Netherlands. Data collection by parent-completed postal questionnaire after telephone contact.	2	4	59	105
	Controls: randomly selected from municipal registers. Data collection by postal questionnaire asking about usual sleeping position in each of months 1–6. Data for month 3 used in analyses.	2	3	39	566
Mitchell 1 1991[97]	1987–1988. SIDS: deaths within areas covering 80% of births in New Zealand.	1		73	128
	Controls: randomly selected in proportion to hospital births in same areas and frequency matched for predicted age and season of cases. Home interviews for both groups measuring position placed at nominated sleep.	1		43	503
Dwyer 1 1991[45]	1988–1990. SIDS: prospective cohort study of births in highest scoring quintile of risk score for SIDS in Tasmania.	2	0	60	15
	Controls: whole cohort excluding SIDS victims. Usual sleeping position prospectively recorded in SIDS and controls at 1 month of age.	2	5	33	2534

Table 2 continued

Author, year published	Study period, selection of cases and controls, and method of data collection	Position recorded[d]	Back (%)	Front (%)	Total studied
Wigfield 1992[90]	1989–1990. SIDS: all deaths in Avon, UK after local publicity about adverse effects of front sleeping.	1		81	32
	Controls: controls selected from same health visitor list as SIDS victim, matched by age and area.	1		28	64
	Data collection by home interview within days of death in both groups.				
Ponsonby 1993[35]	1988–1991. SIDS: all SIDS in Tasmania eligible for inclusion.	2		67	58
	Controls: for each case one control matched for age, and one matched for age and birth weight.	2		30	119
	Data collection by home interviews for both cases and controls. Matched analyses used.				
Jorch 1994[98]	1990–1992. SIDS: cases in two districts in Germany.	1	9	74	94
	Data collected at home interview. Controls: postal survey in two districts of representative sample in Autumn 1991.	1	25	32	758
Gormally 1994[99]	Study period not stated. SIDS: cases identified by the Sudden Infant Death Association in Ireland. Position recorded (usual, put down, found) not stated.	NK	9	79	97
	Controls: matched for sex and age from Rotunda Hospital records in Dublin.	NK	27	26	98
	Data collection by postal questionnaires in both groups.				
Anderson 1995[100]	1984–1992. SIDS: 58% of all SIDS in eastern Norway were enrolled in the study. Parents were sent a postal questionnaire after adverse publicity about front position in 1993. Asked about usual sleeping position between week 2 and death.	2		78	143
	Controls: age, sex, and time matched from birth registry. Postal questionnaire survey conducted in 1993 after adverse publicity about front position. Parents were asked about usual sleeping position for their infants up to 9 years ago between the age of 2 week and death for SIDS, and 2 weeks and 1 year for controls.	2		50	373
Markestad 1 1995[36]	1987–1989. SIDS: cases in county of Hordaland (comprises 10% of births in Norway).	1		78	40
	Controls: randomly selected for another study before 1990. Postal questionnaires sent to case and control parents.	1		64	192
Klonoff–Cohen 1995[37]	1989–1992. SIDS: cases in five health departments in southern California.	2	7	67	193
	Controls: matched by birth date, hospital of birth, sex, and race.	2	10	68	190
	Data collection in both groups by telephone interview before adverse publicity about sleeping position. Control interviews conducted 3–6 months after case interviews.				
[b]					
Taylor 1996[38]	1992–1994. SIDS: cases were residents in King County, USA. Medical examiners asked parents standard questions about sleep position within 48 hours of death.	2		57	47
	Controls: randomly selected using birth certificates for babies born on same date as case.	2	36	25	142
	Data about usual position in previous 2 weeks collected by telephone interview.				
Fleming 2 1996[39]	1993–1995. SIDS: deaths in three English regions.	1	44	16	188
	Controls: matched by age and area from same health visitor list as case.	1	66	3	774
	Data collected at home interview in both groups.				
Mitchell 3 1997[101]	1991–1993. SIDS: cases were all post-neonatal SIDS in New Zealand.	2	10	13	63
	Controls: randomly selected to be representative of all births.	2	24	3	771
	Data for both groups was extracted from routine records recorded by plunket nurses at initial contact and at ~2 months of age				
Brooke 1997[42]	1992–1995. SIDS: all SIDS in Scotland were eligible.	2	31	9	133
	Controls: matched for age, time, and same maternity unit.	2	57	2	256
	Data collected at home visits for both groups.				
Oyen 1997[44]	1992–1995. SIDS: all cases in Norway, Denmark, and Sweden were eligible.	1	13	54	238
	Controls: matched for age, sex, same maternity ward, and time. Parents of both groups contacted soon after death of case. Unclear whether interview or postal survey.	1	44	20	856
Schellscheidt 1997[98,102]	1993–1994. SIDS: cases in two districts in Germany. Follow-on study from Jorch 94.[98]	1	23	59	56
	Controls: selected randomly from same paediatrician as cared for SIDS victim, matched for age and sex. Selection repeated if no response.	1	39	11	156
	Data collection in both groups at home interview.				
Kleeman 1998[103]	1986–1992. SIDS: identified by autopsy in Lower Saxony, Germany.	3		86	140
	Data collection by structured interview with parents.				
	Controls: selected from population register same region.				
	Data collection by postal survey.	3		51	688
Skadberg 1998[36,104]	1990–1995. SIDS: cases in county of Hordaland (10% of births in Norway). Parents asked to complete questionnaire within weeks of death.	1		58	26
	Controls: selected as 10th birth in the county. Data collected by postal questionnaire.	1		5	616

Table 2 continued

Author, year published	Study period, selection of cases and controls, and method of data collection	Position recorded[d]	Back (%)	Front (%)	Total studied
L'Hoir 1998[105]	1995–1996. SIDS: cases in The Netherlands 1995–96 (part of ECAS[41]).	1	53	23	71
	Controls: selected from municipal register or from birth list of nearest large urban hospital, therefore matched for age and area. Data collection at home interview in both cases and controls.	1	87	5	143
Mitchell 2 1999[106]	1987–1990. SIDS: continuation of all New Zealand case–control study using same methods. Contains data from Mitchell 1.[97]	1	5	64	388
	Controls: as previously reported in Mitchell 1.[97]	1	16	33	1584
					4
Dwyer 2 1999[46]	1988–1995. SIDS: cohort study in Tasmania, Australia, measuring usual sleeping position at 1 month of age.	2	3	37	37
	Controls: comparison cohort selected as highest scoring quintile using at risk score for SIDS. Includes data from Dwyer 1 1991[107].	2	6	14	9655
Toro 2001[108]	1996–1998. SIDS: cases from forensic department in Budapest, Hungary, interviewed after autopsy.	NK		61	18
	Controls: controls from primary care units in one district of Budapest, interviewed at regular health checks. No information on how selected but all healthy. Unknown which sleeping position recorded (usual, last placed, or found).	NK		55	74
Hauck 2002[40]	1993–1996. SIDS: cases were Chicago residents (USA).	1	22	57	258
	Controls: selected from the Chicago birth registry matched for maternal age, child's age, and birth weight. Groups of 20–40 controls selected and those responding first included. Data collection at home interview for both cases and controls.	1	33	35	260
McGarvey 2003[43]	1994–1998. SIDS: all cases reported to National SIDS register in Ireland.	1	57	9	203
	Controls: 4 controls randomly selected from birth register matched for geographical location and date of birth. Data collection by home interview within 6 weeks of death or enrolment.	1	61	2	622
Carpenter 2004[41c]	1992–1996. SIDS: studies from 20 regions in Europe. Agreed definitions and pathology investigations and same questions on sleeping position. Cases prospectively identified between 1992–96. Only centres not previously published included in analyses[39,43,44,102,105]	1	25	46	106
	Controls: 2 or more controls selected from birth registers or clinic lists to represent live infants of same age, in same area at the time. All analyses adjusted for age and study using unconditional logistic regression. Data collection by interview within median time of 3 weeks for cases and controls.	1	39	19	228

NK; not known.

[a] The results for side, back and front positions were published in 1972 as a histogram.[89] Actual figures have been supplied by the author.

[b] Studies after this point included populations advised to avoid front sleeping. Four studies were excluded[67,109–111]

[c] Data for separate centres provided by the author and then pooled.

[d] 1 = position placed to sleep before death or interview; 2 = usual position; 3 = position found.

Figure 2d). Recursive meta-analysis showed that the relative magnitude of the cumulative OR for front vs back changed by up to 22% from one publication year to the next between 1986 and 1996, but remained stable (maximum change 4%) when studies published after 1996 were included (results not shown). After 1996, populations included in these studies were advised to use the side or back positions (see Table 2).

Substantial heterogeneity was detected in all three datasets as shown in the forest plots (Figures 2a, c, and e) and reflected in the highly significant Q statistic and high values for I^2 (83% for front vs back, 89% for front vs non-front, and 73% for side vs back). In the conventional meta-regression the only significant factor was the year of recruitment, with later studies associated with an increased OR for SIDS in all three comparisons. The results of extending the meta-regression to include the prevalence of front or side positions in control babies are shown in Table 3. For front compared with back, the prevalence of the front position was the only factor that was significantly associated with heterogeneity. As the prevalence of the front position in control babies increased, the OR for SIDS decreased. For front vs non-front positions, there was little evidence that prevalence of front position or year of recruitment explained heterogeneity. Finally, in the comparison of side vs back, only the prevalence of the side position was associated with a reduction in the OR, but had little effect on heterogeneity.

Discussion

The front sleeping position was recommended from 1943 to 1988 although the first text to advise against front sleeping was not published until 1992. The safest position—on the back—was recommended sporadically during the 1980s but not consistently until 1995. However, by 1970 the pooled evidence from two studies showed that the risk of SIDS was statistically significantly higher for babies on the front than on the back. The harmful effect of front sleeping was lowest when the prevalence of the front position in control babies was highest.

A detailed historical analysis of why clinicians recommended that infants sleep on the front is beyond the scope of this study.

From the reasons given for advocating front sleeping,[47] there is no clear evidence that the back position increases the risk of crying,[46,48–50] inhalation of vomit, or colic.[46,50,51] However, in the short term, sleeping on the front is associated with increased motor development,[52,53] rounder head shape,[54] nappy rash,[49,50] and pyloric stenosis.[55] Front sleeping is also associated with longer sleep duration,[46,48,50] probably by reducing physiological control of respiratory, cardiovascular and autonomic control mechanisms, and arousal during sleep.[56]

Our analyses identified five factors that may have contributed to the delayed recognition of the risks of front sleeping: the paucity of published studies between 1970 and 1986; the marked heterogeneity among studies; the relationship between the prevalence of front sleeping and year of recruitment and the size of the OR; and grouping of the comparator as non-front in some studies. Finally, many authors interpreted the front position as just one of a number of factors associated with SIDS and did not systematically review results from previous

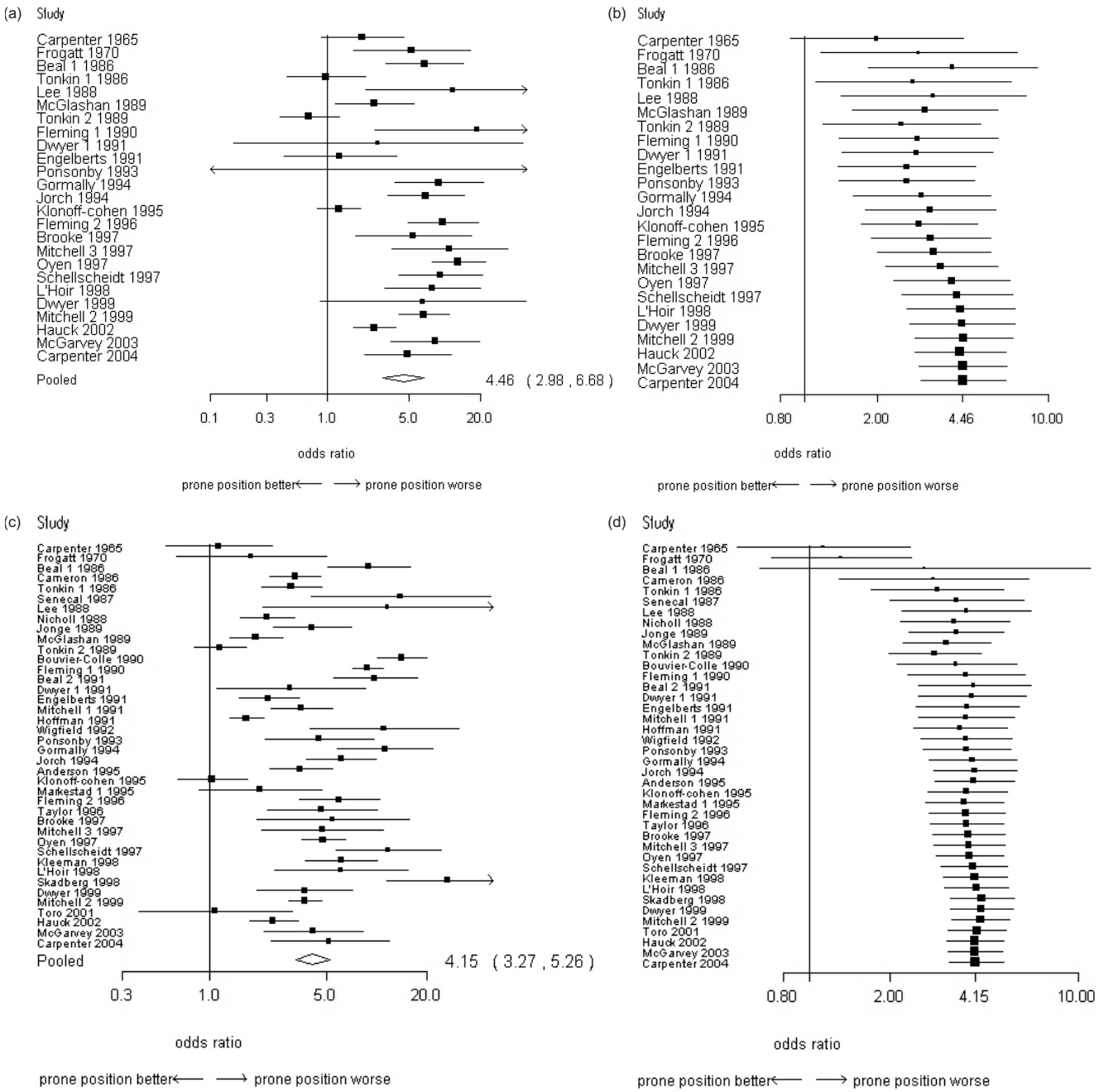

Figure 2 Forest plots show ORs for SIDS and pooled OR for comparisons of (a) front vs back; (c) front vs non-front; and (e) side vs back sleeping positions. Figures 2b, d, and f, depict the cumulative meta-analyses for front vs back (b), front vs non-front (d), and side vs back (f). Studies ordered by publication date

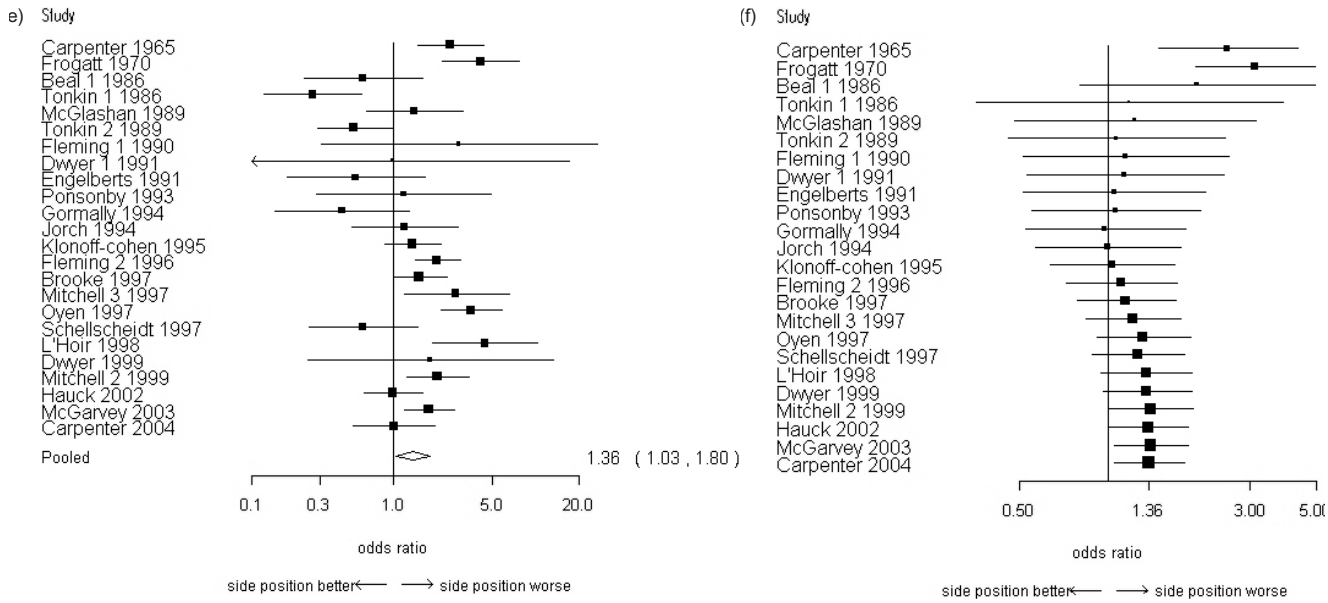

Figure 2 continued

Table 3 Results of meta-regression adjusted for prevalence of front or side position in control infants[a]

Parameter	Adjusted measure of effect (OR)	95% credibility interval	Heterogeneity (95% credibility interval[b])	Change in heterogeneity parameter[c]
Front vs back				
Front vs back	4.92	3.62–6.58	0.31 (0.10–0.71)	0.47
Prevalence of front position	0.75	0.64–0.87		
Midpoint of recruitment period	1.03	1.00–1.07		
Front vs non-front				
Front vs non-front	4.30	3.39–5.39	0.40 (0.21–0.71)	0.07
Prevalence of front position	0.84	0.71–1.00		
Midpoint of recruitment period	1.04	1.01–1.07		
Side vs back				
Side vs back	1.40	3.62–1.84	0.31 (0.11–0.70)	0.002
Prevalence of side position	0.69	0.54–0.89		

[a] Restricted to 38 case–control studies.

[b] Credibility interval: there is a 95% probability that the true value lies within the 95% credibility interval.

[c] Absolute reduction in between-study variance between the crude meta-analysis model and the meta-regression model.

studies.[12,57–59] The first overview of studies on the effect of sleeping position was published by Beal in 1988.[60]

It was striking that no studies were published on the effect of sleeping position between 1970 and 1986. Although several investigators collected data on sleeping position during the 1970s and early 1980s, their findings were not published until 1986 or later.[33,58,59,61–65] Sleeping position may have been disregarded because the front position was not directly compared with the back, and the results of Frogatt and Carpenter were not combined. In addition, Frogatt[66] questioned the validity of his results because they were only statistically significant when the usual sleeping position was compared, not if the position in which the SIDS victim was found was used. Bergman,[67] may have further deterred research on sleeping position after finding that 85% of SIDS victims in a large US study were found on the front, and claiming, without reporting any control data, that this was similar to the community.

The lack of research attention on infant sleeping position between 1970 and 1986 contrasts with the increasing incidence of SIDS, and the steep increase in the proportion of infants sleeping front in several industrialized countries (Figures 3a, b, and c, and Figure 4).[68–75] In the UK, the increase in SIDS incidence was attributed to diagnostic transfer—deaths previously classified as due to respiratory causes being classified

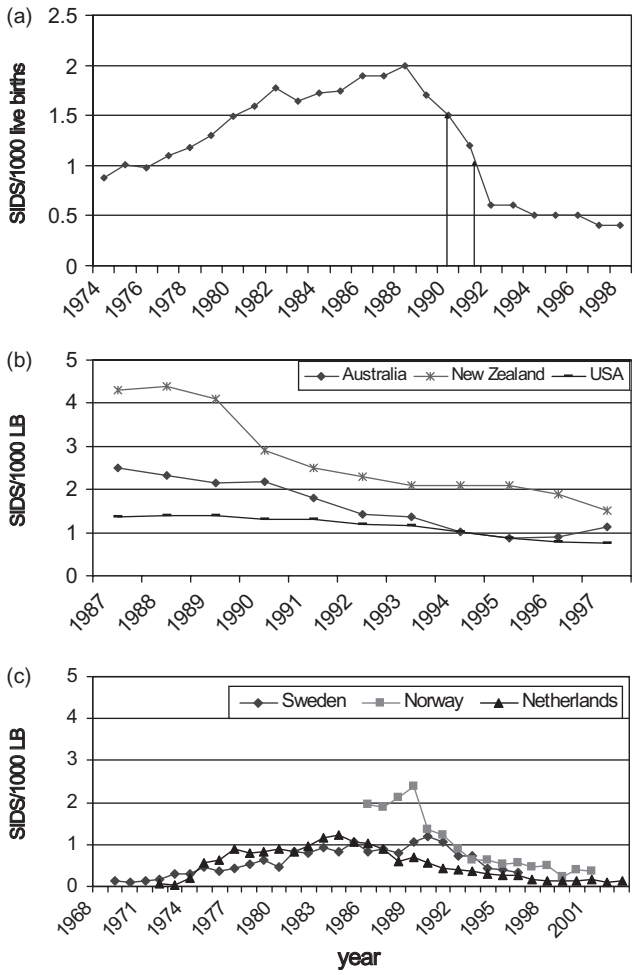

Figure 3 (a) Post-neonatal SIDS mortality (infant deaths due to SIDS after the first month of life) in England and Wales 1974–1998 (Arrows depict publication of Avon SIDS study July 1990,[34] and UK National 'Back to Sleep' campaign, November 1991); (b) SIDS incidence (deaths in the first year per 1000 live births) in Australia, New Zealand and the USA; and (c) SIDS incidence in Sweden, Norway, and The Netherlands

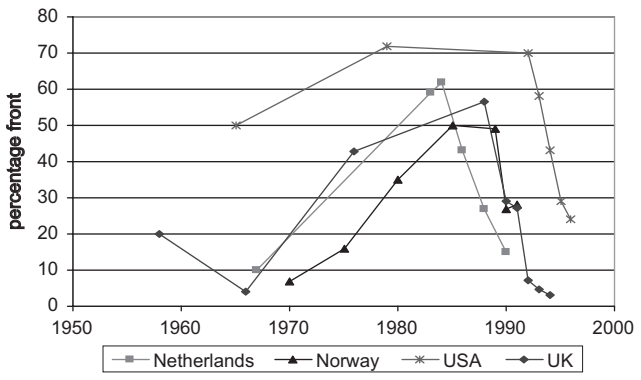

Figure 4 Prevalence of the front position among healthy infants based on controls in included studies and community studies from 1958 to 1998[33,34,39,44,57,63,67–69,84,89–92]

as SIDS, which became a registrable cause only in 1971. However, there was concern that, while all other causes of infant deaths had declined during the 1970s and 1980s, SIDS and respiratory deaths combined had remained static.[75,76] Clear evidence that SIDS incidence had truly increased and was not due to diagnostic transfer was not published until the 1990s (Figure 3c).[68,69,74] In contrast, the decline in incidence following advice to avoid front sleeping in the 'Back to Sleep' campaigns (Figures 3a, b, and c) was rapid and undeniable, providing the strongest evidence to date for a harmful effect of the front position. SIDS incidence fell by 50–70% in numerous countries, in association with a fall in front sleeping. (Figures 3a, b, and c)[75,77,78]

A crude estimate of the number of babies who died in England and Wales owing to harmful health advice can be made by assuming that the rate of post neonatal SIDS would have remained at 0.6/1000 live births, the rate in the year after the government's 'Back to Sleep' campaign. From 1974, when SIDS was routinely used as a cause of death, until 1991, there were 11 000 excess deaths, or nearly 12 extra babies dying each week. However, the number of excess deaths is highest in the USA, where the prevalence of front sleeping was higher for longer than in any other country[48,79](Figure 4). In the USA, rest of Europe, and Australasia, at least 50 000 excess deaths were attributable to harmful health advice.

We found substantial heterogeneity in the association between sleeping position and SIDS that was partly explained by the prevalence of the front (or side) position in control infants, and to a lesser extent, year of recruitment. In an era when front sleeping was the norm, parents who placed infants on the back were likely to have had socioeconomic characteristics that put them at high risk for SIDS, thereby diminishing the observed protective effect of the back position.[80,81] Conversely, when prevailing advice was to avoid front sleeping, characteristics in those that did not take up this advice exaggerated the observed harmful effect of the front position. In other words, increased uptake of advice by families otherwise at low risk of SIDS produced a 'healthy adopter' effect that diminished evidence of harm. An alternative explanation is biased reporting of the position considered to be harmful by parents of SIDS victims. Another possibility is that studies showing an adverse effect of the sleeping position advocated at the time were less likely to be written about and published.

The effect of the era of health advice is best illustrated by comparing the pooled ORs for front vs back positions, before and after advice changed. For studies published between 1965[12] and 1995,[37] the pooled OR was 2.95 (95% CI: 1.69–5.15, studies); thereafter the pooled OR was 6.91 (4.63–10.32). In the example of SIDS, a statistically significant association was still detectable because the OR was relatively large. However, these findings raise a general message for the evaluation of potentially harmful health advice that uptake by people at low risk of adverse outcomes could completely obscure evidence of harm.[82]

The fact that much heterogeneity between studies remained unexplained may be partly owing to difficulties in accurately measuring study characteristics. For example, we could not adequately measure the potential for reporting bias, which may have contributed to the relatively low OR for SIDS in three studies because staff responsible for recommending the front

position also selected control babies and/or collected the data.[61,64,65,83] A second factor in three studies, all conducted in the USA, may be the close matching of controls with cases based on age, hospital of birth, and ethnic group.[37,40,84] If there had been uniform adoption of health advice within these communities, such close matching may have biased the association towards the null effect. In the first two of these studies, close matching, combined with the high prevalence of front sleeping, may have contributed to the relatively weak associations observed.[37,84] Factors contributing to heterogeneity may also differ according to the era of health advice. This may partly explain differences between our results and a previous meta-analysis, restricted to studies published before 1990, that found that country of study, date of publication, matching, and position reported were associated with heterogeneity when sleeping front was compared with non-front.[21]

Conclusions

It is unusual for health advice to have such a profound effect on mortality and to detect such tragic effects from health advice that is not based on evidence of effectiveness. Had systematic reviews been common practice in the early 1970s, parents, professionals, and policy makers would have been aware of the cumulative effect of the front position on SIDS at least 15 years earlier than they were in 1988. Even if the results had been judged insufficient to change practice, they should have stimulated earlier publication of further studies.

Others have similarly highlighted the delayed introduction of effective treatment that could have been avoided if systematic review and meta-analysis had been used to summarize the accumulated evidence from randomized controlled trials.[85,86] Interpretation of systematic reviews of observational studies is more difficult owing to the potential for bias and spurious precision.[87,88] In particular, our results show that observational studies of health advice can be confounded by a 'healthy adopter' phenomenon that can diminish or obscure adverse effects of harmful health advice. All these problems are compounded when examining multiple risk factors. Nevertheless, when randomized controlled trials are lacking or not feasible, systematic review of observational studies is essential to guide policy and practice.

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Conflict of interest

R.G. coordinated one of the included studies.[34] None of the other authors have any conflict of interest.

KEY MESSAGES

- Advice to put infants to sleep on the front for nearly a half century was contrary to evidence available from 1970 that this was likely to be harmful.
- Systematic review of preventable risk factors for SIDS from 1970 would have led to earlier recognition of the risks of sleeping on the front and might have prevented over 10 000 infant deaths in the UK and at least 50 000 in Europe, the USA, and Australasia.
- Attenuation of the observed harm with increased adoption of the front position probably reflects a 'healthy adopter' phenomenon in that families at low risk of SIDS were more likely to adhere to prevailing health advice.

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Appendix: Hierarchical Bayesian model for case-control studies (front vs non-front)

Consider the data in the following 2 × 2 table

	Controls	**Cases**
Front	Fcont	Fcases
Non-front	NFcont	NFcases
	ncont	*ncases*

The number of children sleeping front in cases and controls follow a binomial distribution (B) with probabilities $P(\text{front}|\text{control})$ and $P(\text{front}|\text{cases})$

$$\text{Fcont} \sim \text{B}(P(\text{front}|\text{control}), n\text{cont})$$
$$\text{Fcases} \sim \text{B}(P(\text{front}|\text{case}), n\text{cases})$$

Then we parameterize as

$$\text{logit}(P(\text{front}|\text{control})) = u$$
$$\text{logit}(P(\text{front}|\text{cases})) = u + \text{logOR}$$

then, logOR is regressed as

$$\text{logOR} = a + \beta_1 u + \beta_2 D$$

where now the intercept a is the adjusted log OR, D is the mean date of recruitment, and β_1 is the coefficient for the dependence of the OR on the prevalence of front sleeping.