Swaddling and the Risk of Sudden Infant Death Syndrome: A Meta-analysis

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abstract

CONTEXT: Swaddling is a traditional practice of wrapping infants to promote calming and sleep. Although the benefits and risks of swaddling in general have been studied, the practice in relation to sudden infant death syndrome remains unclear.

OBJECTIVE: The goal of this study was to conduct an individual-level meta-analysis of sudden infant death syndrome risk for infants swaddled for sleep.

DATA SOURCES: Additional data on sleeping position and age were provided by authors of included studies.

STUDY SELECTION: Observational studies that measured swaddling for the last or reference sleep were included.

DATA EXTRACTION: Of 283 articles screened, 4 studies met the inclusion criteria.

RESULTS: There was significant heterogeneity among studies (I^2 = 65.5%; P = .03), and a random effects model was therefore used for analysis. The overall age-adjusted pooled odds ratio (OR) for swaddling in all 4 studies was 1.58 (95% confidence interval [CI], 0.97–2.58). Removing the most recent study conducted in the United Kingdom reduced the heterogeneity (I^2 = 28.2%; P = .25) and provided a pooled OR (using a fixed effects model) of 1.38 (95% CI, 1.05–1.80). Swaddling risk varied according to position placed for sleep; the risk was highest for prone sleeping (OR, 12.99 [95% CI, 4.14–40.77]), followed by side sleeping (OR, 3.16 [95% CI, 2.08–4.81]) and supine sleeping (OR, 1.93 [95% CI, 1.27–2.93]). Limited evidence suggested swaddling risk increased with infant age and was associated with a twofold risk for infants aged >6 months.

LIMITATIONS: Heterogeneity among the few studies available, imprecise definitions of swaddling, and difficulties controlling for further known risks make interpretation difficult.

CONCLUSIONS: Current advice to avoid front or side positions for sleep especially applies to infants who are swaddled. Consideration should be given to an age after which swaddling should be discouraged.

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Ms Pease and Dr Blair conceptualized and designed the review; conducted the search, screening, and analyses; and wrote the initial draft of the paper. Drs Fleming, Hauck, and Ponsonby provided additional individual-level data for the analysis, and reviewed and revised the manuscript. Drs Moon, L'Hoir, and Horne provided

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Swaddling is defined as close wrapping of an infant, usually with a light cloth and the head exposed, although swaddling styles vary across cultures. It has been reported that swaddling infants for sleep is gaining popularity in countries such as the United Kingdom and the United States,[1–3] with several reported benefits to the practice. For the general population of young infants, these include more quiet sleep,[3] fewer spontaneous arousals during quiet sleep,[4] and a slight reduction in excessive crying in infants aged <7 weeks.[5] Further benefits are observed for specific populations, including improvements in neuromuscular development for very low birth weight infants,[6] reduced physiologic and behavioral distress among premature infants,[7] and improved calming and sleep for infants with neonatal abstinence syndrome.[8]

Impaired cardiovascular control in association with a failure to arouse from sleep, leading to a reduced ability to respond to a cardiovascular stress (eg, profound hypotension), has been described as a potential mechanism leading to some sudden infant death syndrome (SIDS) deaths.[9] It has also been proposed that, due to the effects of swaddling on sleep, swaddling could be used as an intervention to support supine sleep for parents who report that their infant has difficulty sleeping in this position.[10] There is some evidence that infants who are swaddled and sleep supine exhibit greater heart rate responses to an auditory stimulus than infants sleeping supine nonswaddled.[11] In contrast, there is evidence that swaddling may increase the risk of SIDS, as swaddled infants have fewer spontaneous arousals from sleep and increased sleep time, particularly during quiet sleep, which is a state of reduced arousability.[4] In addition, those infants who were naive to swaddling (ie, not routinely swaddled) had higher induced arousal thresholds in both sleep states[2] and reduced spontaneous arousability in active sleep.[12] Beyond the field of SIDS, swaddling may also increase an infant's risk for developmental dysplasia of the hip (especially if not applied well),[13] hyperthermia,[1] pneumonia, and upper respiratory tract infections.[14]

To date, the association between swaddling and SIDS risk remains unclear, and to the best of our knowledge, no comprehensive review of the literature has been performed to determine the strength of this relationship. Although health professionals and parents need this information to help make safe infant care choices, currently no official guidance exists in the United Kingdom or the United States on this topic. The purpose of the present investigation was to conduct a systematic review of the literature on this issue and quantify, through meta-analysis and using individual data when possible, whether swaddling is associated with an increased risk of SIDS.

METHODS

Search Strategy

A thorough literature search was conducted to identify studies published from the 1950s to December 2014. A comprehensive search of online databases was conducted on 3 platforms (PubMed, Web of Science, and Ovid SP) and included Medline, AMED (Allied and Complementary Medicine), CAB Abstracts, Embase, PsycINFO, BIOSIS Citation Index, SciELO Citation Index, and Web of Science Core Collection. The following search terms were used: SIDS, SUDI, SUID, swadd* (to include swaddling, swaddled, and swaddle), wrap* (to include wrapping, wrapped), and swath* (to include swathe, swathing, and swathed). Swaddling, wrapping, and swathing are used alternately and may refer to different methods of swaddling in different countries; in general, these terms are used to describe the practice of wrapping an infant in blankets or cloth. In addition, the authors conducted secondary hand-searches of reference lists of all articles found in the online database search and requested unpublished data at national and international SIDS conferences.

Selection of Eligible Studies

The online database search returned 389 results, and additional data from hand-searches and unpublished data (other sources) returned 15 records; after removing duplicates, this total was reduced to 283. The initial screen of titles and abstracts excluded 272 records, and the remaining 11 full-text records were assessed. Studies were excluded if they were not case-control or cohort by design, if they did not use SIDS as an outcome, or if they did not mention swaddling or infant wrapping. Articles in languages other than English had the abstract translated for screening. There were 2 studies in which it was unclear if wrapping was the same as swaddling; in both cases, the investigators were contacted to clarify the definition. Four studies were deemed eligible for inclusion in this review,[15–18] including 1 case-control study[18] in which the swaddling data were collected but remained unpublished. The screening process is detailed in Fig 1.

Data Extraction and Synthesis

With only 4 studies included in the review, data were extracted by hand individually from each article or source. The investigators were contacted where necessary to provide further data on postnatal age (in days) and the sleeping position (supine, side, or prone) in which swaddled and nonswaddled infants were placed (both control

subjects and infants with SIDS) for their last sleep and were found. The Newcastle-Ottowa scale was used to assess study quality.[19,20]

Statistical Methods

Descriptive statistics included median and interquartile ranges (IQRs) for data that were not normally distributed. The χ^2 test was used to compare proportional differences. The Mann-Whitney *U* test was used for continuous unpaired data. The final sleep of the SIDS case subjects was compared with the reference sleep of age-matched (on an individual basis) surviving control infants for 3 of the studies and age-weighted (on a group basis) surviving control subjects for 1 study. Odds ratios (ORs) with 95% confidence intervals (CIs) were calculated by using χ^2 tests. Logistic regression was used to adjust the findings for age. A meta-analysis was conducted with the use of a fixed effects model and a random effects model in which heterogeneity proved significant. The latter allows for heterogeneity by assuming that the true effect varies between studies. It provides a more conservative pooled estimate reflecting the greater uncertainty when using heterogeneous data. The strength of the association between swaddling and SIDS for each study was illustrated by using a forest plot. Data were pooled and analyzed by using Stata version 13.1 (Stata Corp, College Station, TX).

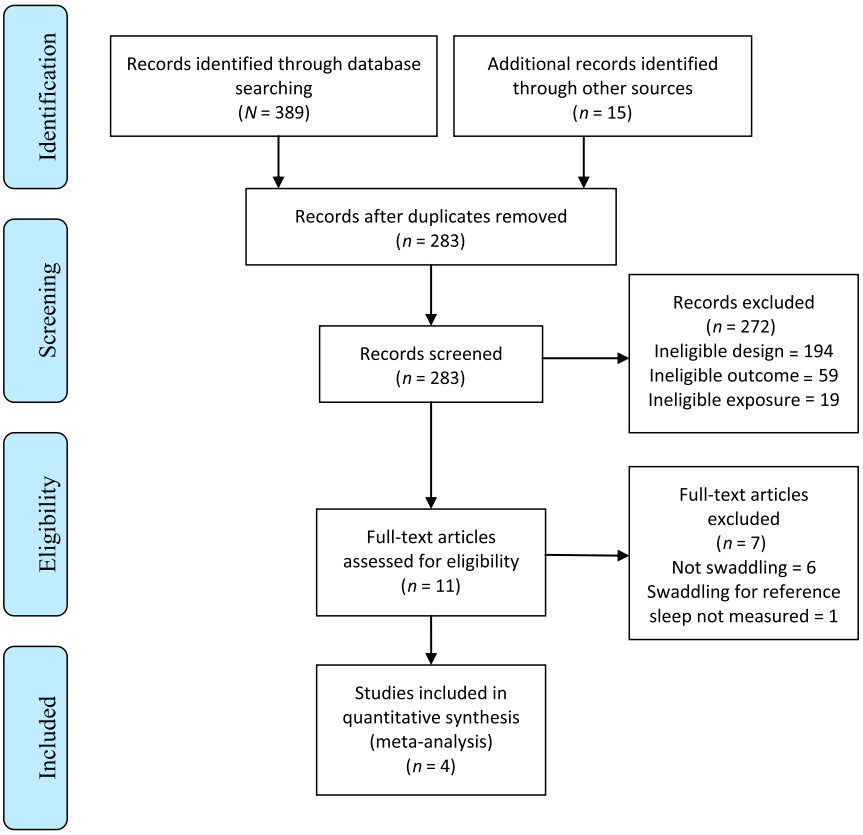

FIGURE 1
Flowchart of study selection based on inclusion and exclusion criteria.

RESULTS

Included Studies and Swaddling Prevalence

A total of 4 case-control studies that examined the association between swaddling and SIDS were included in the review, and the study characteristics are summarized in Table 1. The 4 studies span 2 decades and 3 diverse areas: regions of England in the United Kingdom, Tasmania in Australia, and Chicago, Illinois, in the United States. None of the studies gave a precise definition of swaddling. Prevalence of swaddling during the different time periods varied in the 3 areas suggested by the control populations. Prevalence was highest in the study from Tasmania in the late 1980s (35.7%) and much lower in Chicago in the mid-1990s (9.2%) and England (10.3%), with a slight decrease in England 10 years later (5.7%); the number of infants included in this latter study was small (only 87 control subjects). Across the studies, 17.5% of the infants with SIDS were swaddled for the last sleep compared with 10.8% of similar-aged surviving control infants during a designated reference sleep. In all 4 included studies, the proportion of infants swaddled was higher in the SIDS cases than in the control cases; however, only in the 2 UK studies[15,18] was the univariate association significant, and only in the more recent of these studies[15] did this significance remain after adjustment for other factors. The assessment of study quality indicated that these were good-quality observational studies (Table 2). All 4 studies used independent case definitions and consecutive case recruitment methods. They all used community control subjects and considered comparability in the design and analysis stages. None of the studies blinded the interviewer to case or control status (although this approach would have been unethical given the outcome under study). Nonresponse differences between case and control subjects were reported in 2 of the 4 studies, and the potential for bias was raised as a limitation.

TABLE 1 Study Characteristics

Source	Study Area	Period	SIDS Defined	Swaddling Defined	Control Group Selection
Ponsonby et al[16,21]	Tasmania, Australia	1988–1991	After postmortem by hospital pathologist, including toxicologic and bacteriologic screenings	Infant wrapped in an item of bedding (eg, sheet or light blanket) while asleep	96% aged within 7 d of each case subject, 100% within 14 d
Fleming et al[18,22–24]	England, UK	1993–1996	Using the Avon Clinico-Pathological Classification System and 1989 definition of SIDS[25]	Infant wrapped in an item of bedding (eg, sheet or light blanket) while asleep	65.8% aged within 14 d of each case subject, 91.2% within 1 mo
Hauck et al[17,26]	Chicago, Illinois, USA	1993–1996	1989 definition of SIDS[25]	Wrapped/swaddled	88% aged within 14 d of each case subject, 100% within 1 mo; matched on maternal race and birth weight
Blair et al[15]	Southwest England, UK	2003–2006	Using the Avon Clinico-Pathological Classification System and 1989 definition of SIDS[25]	Infant wrapped in an item of bedding (eg, sheet or light blanket) while asleep	Weighted distribution to mimic age distribution of earlier case–control SIDS case subjects, adjusted to the age distribution of SIDS in the present study sample

TABLE 2 Study Quality Assessments Using the Newcastle-Ottowa Scale

Variable	Study			
	Ponsonby et al,[16] 1988–1991	Fleming et al,[18] 1993–1996	Hauck et al,[17] 1993–1996	Blair et al,[15] 2003–2006
Selection				
Is the case definition adequate (with independent validation)?	Yes, after postmortem examination (1*)	Yes, using the Avon Clinico-Pathological Classification System (1*)	Yes, with independent validation, as determined by the Office of the Medical Examiner (1*)	Yes, using the Avon Clinico-Pathological Classification System (1*)
Representativeness of the case subjects	Consecutive series using professional state ambulance service covering 94% of the population (1*)	Consecutive series using established notification network system (1*)	Consecutive series using Office of the Medical examiner notification system (1*)	Consecutive series using established notification network system (1*)
Selection of control subjects	Community control subjects, from maternity lists (1*)	Community control subjects, from health visiting lists (1*)	Community control subjects, from ongoing review of birth certificates (1*)	Community control subjects from maternity wards, weighted for selection bias according to maternal social class (1*)
Definition of control subjects	No history of disease (infant has not died) (1*)	No history of disease (infant has not died) (1*)	No history of disease (infant has not died) (1*)	No history of disease (infant has not died) (1*)
Comparability				
Comparability of case subjects and control subjects on the basis of the design or analysis	(1) Study controls for infant age (1*); (2) Study controls for additional factors (time of last or reference sleep) (1*)	(1) Study controls for infant age (1*); (2) Study controls for additional factors (time of last or reference sleep) (1*)	(1) Study controls for infant age (1*); (2) Study controls for additional factors (maternal ethnicity, infant birth weight, time of last or reference sleep) (1*)	(1) Study controls for infant age (1*); (2) Study controls for additional factors (random control subjects matched for social class) (1*)
Exposure				
Ascertainment of exposure	Interviewer not blinded to case/control status	Interviewer not blinded to case/control status	Interviewer not blinded to case/control status	Interviewer not blinded to case/control status
Same method of ascertainment for case infants and control subjects	Yes, semi-structured interviews (1*)	Yes, semi-structured interviews (1*)	Yes, semi-structured interviews (1*)	Yes, semi-structured interviews (1*)
Nonresponse rate	Nonrespondents described: 6% case subjects, 17% control subjects	Same rate for both groups: 8.7% case subjects, 7.5% control subjects (1*)	Nonrespondents described: 0.0% case subjects, 95.5% control subjects	Same rate for both groups: 4% case subjects, 5% random control subjects (1*)
Total stars (of 9)	7/9	8/9	7/9	8/9

TABLE 3 Individual Univariate and Pooled ORs for Swaddling and SIDS

Source	SIDS Cases (Swaddled/*N*)		Control Subjects (Swaddled/*N*)		OR[a]		Adjusted OR (Where Available)	
	n/*N*	%	*n*/*N*	%	OR	95% CI	OR	95% CI
Ponsonby et al[16]	38/103	36.9	40/112	35.7	1.05	0.59–1.87		
Fleming et al[18]	47/319	14.7	119/1300	9.2	1.63	1.13–2.36		
Hauck et al[17]	29/260	11.2	26/260	10.0	1.16	0.66–2.06	1.0[b]	0.5–1.9
Blair et al[15]	19/78	24.4	5/87	5.7	5.82	2.02–16.74	31.1[c]	4.2–228.9
Total	133/760	17.5	190/1759	10.8	1.53	1.18–1.97 (fixed effects)		
					1.58	0.97–2.58 (random effects)		
Excluding Blair et al					1.38	1.05–1.80 (fixed effects)		

[a] The difference in swaddling within each individual study was adjusted for infant age, as were the pooled estimates for both the fixed and random effects models.
[b] Adjusted for maternal age, marital status, education, and index of prenatal care (using the number of prenatal visits and when prenatal care was initiated).
[c] Adjusted for infant age, daytime or nighttime sleep, prematurity, larger families, maternal consumption of >2 Units of alcohol in the last 24 hours, infant bed-sharing with an adult, co-sleeping with an adult on the sofa, infants sleeping outside the parental bedroom, use of pillows, maternal education, smoking during pregnancy, infant found prone, and infant unwell in the last 24 hours.

Pooled Results and Heterogeneity

In total, 760 SIDS cases were compared with 1759 control subjects. The fixed effects model, assuming equal variance, suggests a significant pooled OR of 1.53 (95% CI, 1.18–1.97; $P = .001$). However, the prevalence of swaddling between studies was variable, and an I^2 statistic of 65.5% indicated significant and substantial heterogeneity between studies ($P = .03$). To adjust for the heterogeneity, a random effects model was applied, yielding a pooled OR of borderline significance (1.58 [95% CI, 0.97–2.58]; $P = .06$). Looking more closely at the variance, the studies were homogeneous (I^2 statistic of 28.2%; $P = .25$) when the most recent UK study was excluded. The pooled OR for swaddling during last sleep for the 3 remaining studies, by using a fixed effects model of analysis (shown in Table 3), provided a summary OR that remained significant (OR, 1.38 [95% CI, 1.05–1.80]; $P = .02$). The forest plot for this model is shown in Fig 2.

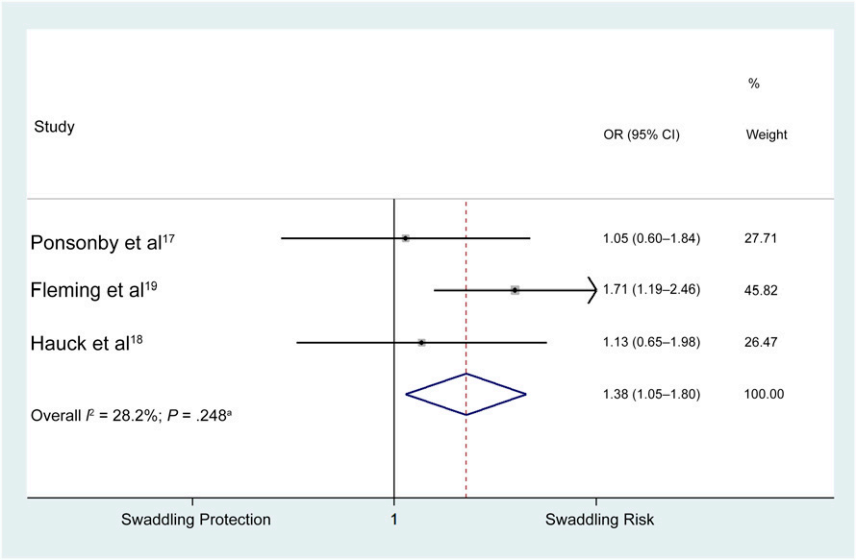

FIGURE 2
Forest plot with 3 of 4 included studies showing the fixed effects pooled result, adjusted for infant age. [a]The pooled result is illustrated by the diamond, the lateral points of which indicate the CIs for the estimate.

Swaddling and Infant Postnatal Age

Swaddled infants tended to be younger (median age, 57 days [IQR, 35–94 days], compared with 96 days [IQR, 60–154 days] for nonswaddled infants in all 4 studies), although there was no significant difference ($P = .23$) between the age of the swaddled SIDS infants (median age, 55 days [IQR, 30–96 days]) and swaddled control infants (median age, 61 days [IQR, 37–94 days]). Figure 3 shows the age distributions of SIDS case and control infants swaddled and not swaddled. Swaddling in all groups peaked at ~2 months of age and dropped off steeply between 4 and 6 months of age. Although the numbers were small, the risk of SIDS from swaddling increased with age (Table 4), with the highest risk associated with infants aged ≥6 months (OR, 2.53 [95% CI, 1.21–5.23]).

Swaddling and Sleeping Position

Data for position in which infants were placed for the last sleep were available from the UK and Chicago studies, whereas data for position in which infants were found were available in the UK and Tasmanian studies. In terms of position placed for the last sleep (Table 5), being swaddled and placed prone conferred the greatest risk, although this practice was rare. For both the prone and side positions, the risk doubled when the infant was also swaddled compared with nonswaddled.

TABLE 4 ORs for Infant Swaddling During Last Sleep Stratified According to Infant Age Categories

Infant Age	Swaddled	SIDS Case Subjects		Control Subjects		OR	95% CI
		n	%	*n*	%		
<1 mo	No	10	47.6	6	54.5	1.00	Ref group
	Yes	11	52.4	5	45.5	1.32	0.31–5.70
1–2 mo	No	215	76.0	471	82.3	1.00	Ref group
	Yes	68	24.0	101	17.7	1.48	1.04–2.09
3–5 mo	No	267	87.0	678	91.0	1.00	Ref group
	Yes	40	13.0	67	9.0	1.52	0.99–2.29
≥6 mo	No	135	90.6	414	96.1	1.00	Ref group
	Yes	14	9.4	17	3.9	2.53	1.21–5.23

TABLE 5 ORs for Infant Swaddling During Last Sleep Stratified According to Position Placed for Sleep

Sleeping Position	Swaddled	SIDS Case Subjects	Control Subjects	OR[a]	95% CI
Supine	No	210 (32.4%)	970 (59.1%)		Ref group
Supine	Yes	38 (5.9%)	85 (5.2%)	1.93	1.27–2.93
Side	No	150 (23.1%)	390 (23.8%)	1.73	1.35–2.19
Side	Yes	44 (6.8%)	61 (3.7%)	3.16	2.08–4.81
Prone	No	195 (30.0%)	131 (8.0%)	6.75	5.16–8.82
Prone	Yes	12 (1.8%)	4 (0.2%)	12.99	4.14–40.77

Analysis combines data from the United Kingdom[15,18] and the United States.[17]
[a] Adjusted for infant age.

TABLE 6 ORs for Infant Swaddling During Last Sleep Stratified According to Position Found

Sleeping Position	Swaddled	SID Case Subjects	Control Subjects	OR[a]	95% CI
Supine	No	147 (30.7%)	1039 (72.1%)		Ref group
Supine	Yes	31 (6.5%)	110 (7.6%)	1.86	1.19–2.88
Side	No	76 (15.9%)	142 (9.9%)	3.61	2.59–5.02
Side	Yes	32 (6.7%)	48 (3.3%)	4.33	2.67–7.02
Prone	No	155 (32.4%)	97 (6.7%)	12.47	9.06–17.17
Prone	Yes	38 (7.9%)	5 (0.4%)	49.86	19.28–128.95

Analysis combines data from the United Kingdom[15,18] and Tasmania, Australia.[16]
[a] Adjusted for infant age.

Notably, there was still a small but significant risk associated with infants being swaddled and placed for sleep on their backs. In terms of position found for the last sleep (Table 6), being swaddled and found sleeping prone was a rare event among the control subjects (<1%) but was more frequent among the infants with SIDS (8%).The numbers were small, but even when the lower CI was used, the risk was 19-fold. Being found on the side while swaddled was a slightly higher risk than being placed on the side without swaddling. Again, being found swaddled on the back conferred a small but significant risk compared with being found on the back nonswaddled.

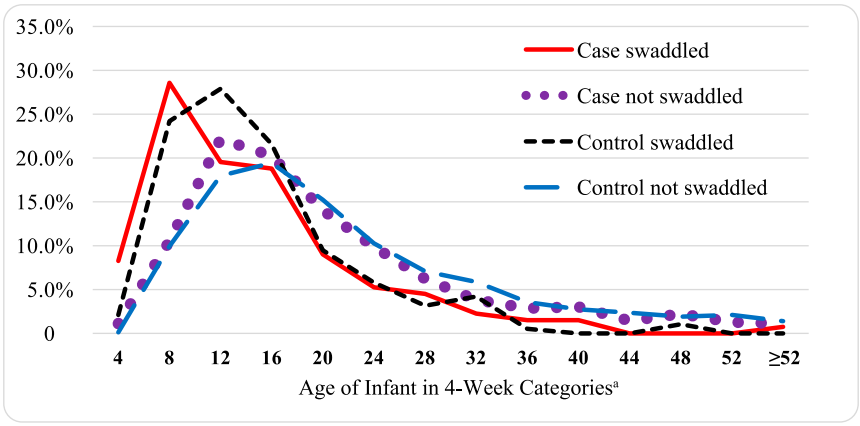

FIGURE 3
Comparison of infant age distribution among SIDS case subjects and control subjects according to swaddling status. [a]Categories in 4-week intervals (eg, 0–28 days, 29–56 days).

Only the 2 UK studies included data on both the sleeping position placed for the last sleep and position found. Of the 124 control infants who were swaddled, none was placed prone, those placed on their side either remained on their side (24 infants) or rolled on to their back (25 infants), and 1 control infant was placed supine and found prone. However,

among the 16 swaddled infants with SIDS found prone, 3 were placed and found prone, 8 were placed on their side and found prone, and the remaining 5 were placed supine and found prone; the mean age of these 5 infants was >15 weeks (compared with 11 weeks among the other swaddled infants in these 2 studies).

DISCUSSION

The present analysis combines case–control data from 4 studies on the risk of SIDS for infants who were swaddled. The results from the individual-level meta-analysis revealed an age-adjusted pooled risk between swaddling and SIDS, although this finding was of borderline significance when the heterogeneity of studies was taken into account. The data extracted were limited to 4 studies from diverse regions, during different time periods, with variable prevalence rates for swaddling. The main source of heterogeneity seemed to originate from the latest study in the United Kingdom; the data for this study were collected at least 10 years after the other studies in this analysis. Although it is possible that this study is idiosyncratic, it may also indicate that interval changes in sleep practices in the United Kingdom have made the practice of swaddling more risky. Future observational studies will be needed to determine this possibility. Swaddling as a risk seemed to vary according to sleep position and older age. The significant risk of placing infants on their side or prone to sleep doubled when infants were swaddled, and the SIDS risk associated with swaddling increased with age.

It has been suggested that swaddling can be used to encourage supine sleep when families are having difficulty settling younger infants in this position.[27,28] Evidence regarding the effectiveness of this strategy is limited, although a survey from Oden et al[10] found that the supine position for sleep was more common in infants who were routinely swaddled than in those who were swaddled occasionally. The limited evidence from the analysis in the present article does not suggest that placing younger swaddled infants supine for sleep has a protective effect against SIDS, although further studies are needed to quantify whether this practice poses any risk. It has also been postulated that swaddling could decrease the risk for SIDS by making the side position more stable.[29] Our evidence suggests this outcome is the case because the risk associated with being placed in the side position almost doubled among swaddled infants. Few infants were swaddled and placed prone, although this practice was significantly more common among the infants who died, and the risk of SIDS from being placed prone doubled among the swaddled infants. We already know that being found prone is strongly associated with SIDS, but this risk also increased fourfold among the swaddled infants. The handful of swaddled SIDS infants for whom we had data suggests that the majority of those found prone either moved into this position after being placed on their side or they were older infants who rolled from the supine to the prone position.

Swaddling was a more common practice among the younger infants, and the risk of SIDS linked to swaddling seemed to increase in older infants. This outcome may be related to a greater likelihood of rolling to a prone position at an older age. The advice given in the Netherlands, a country with low rates of SIDS, is to encourage swaddling to reduce excessive crying.[30] Because of the likelihood of rolling to the prone position, the official advice in the Dutch guideline on excessive crying is never to initiate swaddling after the fourth month, to stop swaddling as soon as the child signals he or she is trying to turn over, and always to stop swaddling before the sixth month (after this age, infants will be able to roll over).

There are several limitations to the present meta-analysis. The heterogeneity between studies was substantial, reflecting the differing swaddling practices across countries and time periods. Due to the differences in control selection, it was not possible to use conditional logistic regression, which may have increased the power of detecting the pooled estimate. The high rates of swaddling in the study from Tasmania may reflect different cultural practices at the time of that particular study. Although the quality of the studies was high, none adequately described the swaddling technique, and the questions used in each study (provided by the investigators) sometimes used the terms wrapping and swaddling synonymously, when they might indicate different infant care practices. Safer forms of swaddling in the supine position have been explored in sleep laboratory studies of young infants, especially those considering normal hip development and chest pressure concerns,[27] and guidelines are more specific in some countries than others. The individual-level analysis adjusted swaddling for infant age and sleeping position but many other factors associated with SIDS were not adjusted for, in particular whether the infant was bed-sharing with the parents.

Given that the most recent (albeit small) case–control study included here found that swaddling was a significant risk in the multivariate analysis, future studies of SIDS risk should include a detailed assessment of swaddling to further understand this infant care practice. Future investigations of swaddling could include photographic measures of how a swaddled child was placed for sleep and found thereafter, which would improve swaddling classifications.

CONCLUSIONS

Despite the limitations, these analyses indicate that the current advice to avoid placing infants on their front or side to sleep may especially apply to infants who are swaddled. Given the marked increase in infants swaddled and found prone (rather than placed prone), coupled with an increased risk of swaddling with increased age regardless of sleeping position, health professionals and current guidelines should consider an appropriate age limit at which swaddling should be discouraged.

ABBREVIATIONS

CI: confidence interval
IQR: interquartile range
OR: odds ratio
SIDS: sudden infant death syndrome

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