Risk Factor Changes for Sudden Infant Death Syndrome After Initiation of Back-to-Sleep Campaign

WHAT'S KNOWN ON THIS SUBJECT: Prone sleep, bed-sharing, maternal smoking during pregnancy, and prematurity increase the risk of sudden infant death syndrome. The sudden infant death syndrome rate initially declined dramatically after the initiation of the US Back-to-Sleep campaign in 1994, but subsequently plateaued.

WHAT THIS STUDY ADDS: The risk profile has changed since the Back-to-Sleep campaign; the prevalence of simultaneous risks has remained consistent. Intrinsic and extrinsic risks provide unification into 1 underlying triple-risk model and insights into potential underlying mechanisms.

abstract

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OBJECTIVE: To test the hypothesis that the profile of sudden infant death syndrome (SIDS) changed after the Back-to-Sleep (BTS) campaign initiation, document prevalence and patterns of multiple risks, and determine the age profile of risk factors.

METHODS: The San Diego SIDS/Sudden Unexplained Death in Childhood Research Project recorded risk factors for 568 SIDS deaths from 1991 to 2008 based upon standardized death scene investigations and autopsies. Risks were divided into intrinsic (eg, male gender) and extrinsic (eg, prone sleep).

RESULTS: Between 1991–1993 and 1996–2008, the percentage of SIDS infants found prone decreased from 84.0% to 48.5% ($P < .001$), bed-sharing increased from 19.2% to 37.9% ($P < .001$), especially among infants <2 months (29.0% vs 63.8%), prematurity rate increased from 20.0% to 29.0% ($P = .05$), whereas symptoms of upper respiratory tract infection decreased from 46.6% to 24.8% ($P < .001$). Ninety-nine percent of SIDS infants had at least 1 risk factor, 57% had at least 2 extrinsic and 1 intrinsic risk factor, and only 5% had no extrinsic risk. The average number of risks per SIDS infant did not change after initiation of the BTS campaign.

CONCLUSIONS: SIDS infants in the BTS era show more variation in risk factors. There was a consistently high prevalence of both intrinsic and especially extrinsic risks both before and during the Back-to-Sleep era. Risk reduction campaigns emphasizing the importance of avoiding multiple and simultaneous SIDS risks are essential to prevent SIDS, including among infants who may already be vulnerable. *Pediatrics* 2012;129:630–638

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KEY WORDS

triple-risk model, brainstem, prone sleep, bed-sharing, serotonin, maternal smoking

ABBREVIATIONS

BTS—Back-to-Sleep
SIDS—sudden infant death syndrome
URTI—upper respiratory tract infection

All authors had full access to all of the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis; Drs Krous and Haas were responsible for study concept and design; Drs Krous, Haas, and Stanley were responsible for acquisition of the data; Drs Trachtenberg, Haas, Krous, and Kinney were responsible for analysis and interpretation of data; Drs Trachtenberg and Krous were responsible for drafting of the manuscript; Drs Krous, Haas, Kinney, and Stanley were responsible for critical revision of the manuscript for important intellectual content; Dr Trachtenberg was responsible for statistical analysis; Drs Krous and Kinney obtained funding; Drs Krous and Haas were responsible for administrative, technical, or material support; and Dr Krous was responsible for study supervision.

www.pediatrics.org/cgi/doi/10.1542/peds.2011-1419

doi:10.1542/peds.2011-1419

Accepted for publication Dec 14, 2011

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PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

FINANCIAL DISCLOSURE: *The authors have indicated they have no financial relationships relevant to this article to disclose.*

FUNDING: This study was supported by the CJ Foundation for SIDS, First Candle/SIDS Alliance, the Southwest SIDS Research Institute, and National Institute of Child Health and Development grant HD-20991 to Dr Kinney.

The major advance in research into sudden infant death syndrome (SIDS) is the recognition that the prone sleep position increases risk twofold or more.[1] The National Institute of Child Health and Development initiated in 1994 the "Back-to-Sleep" (BTS) campaign in the United States that advised caregivers to place infants on their backs to sleep.[1] In 1994, the triple-risk model for SIDS (Fig 1) crystallized in a simple Venn diagram the current thinking: (1) a vulnerable infant with an underlying susceptibility; (2) an exogenous stressor at the time of death; and (3) the critical developmental period, with a peak at 2 to 4 postnatal months.[2] In a spectacular achievement for public health, the SIDS rate in the United States declined by >50% in the 10 years after the initiation of the BTS campaign.[3] The current American Academy of Pediatrics SIDS risk reduction guidelines also include recommendations against side sleep and bed-sharing, and suggest a separate but proximate sleeping environment and pacifier use.[3] Still, the overall SIDS rate has plateaued, and SIDS remains the leading cause of postneonatal infant mortality in the United States today, with an overall incidence of 0.53/1000.[4,5] Thus, the need to understand the precise risk factors for SIDS remains critical today to design timely risk reduction messages, as well as relevant hypotheses for basic research into SIDS pathogenesis. Evidence is mounting from European studies that rates of bed-sharing, poverty, prematurity, and maternal smoking during pregnancy in SIDS have increased, but placing the infant prone to sleep has decreased.[6] Yet, more information is needed in the United States to tailor risk reduction messages specifically to this country. Our objectives in a large US population of well-characterized SIDS infants were to: (1) test the hypothesis that the profile of SIDS risk differs before and after the initiation of the BTS campaign; (2) determine the prevalence and patterns of co-occurrence of multiple risks; and (3) establish the age profile of risk factors.

METHODS

SIDS was defined as the sudden unexpected death of an infant <1 year of age, with onset of the fatal episode apparently occurring during sleep, that remains unexplained after a thorough investigation, including performance of a complete autopsy and review of the circumstances of death and the clinical history.[7] Infant deaths that did not meet these criteria for SIDS, but for which alternative diagnoses of natural or unnatural conditions were equivocal, were diagnosed as unclassified sudden infant death and were excluded from risk factor analysis in this study. This study population comprises 100% of infant deaths from the San Diego Medical Examiner's Office from 1991 to 2008. All SIDS cases were autopsied according to standard protocols. Trained medical examiner investigators collected demographic and risk factor information within 30 hours of death. This study was approved by the Institutional Review Board of Rady's Children's Hospital, San Diego. Parental informed consent for research was not required under the California SIDS Autopsy Statute of 1991 which mandates standardized death scene investigation and autopsy on all cases of sudden unexpected infant death.

Study years were grouped as pre-BTS (1991–1993), the transition period in infant sleep practices in San Diego County (1994–1995), and the BTS era (1996–2008) (ongoing to the present). The postnatal ages of the infants were grouped as <2 postnatal months, 2 to 4 months, and 5+ months, because SIDS historically is most common between 2 and 4 months of age;[3] results were similar when cases were grouped by postconceptional age (postnatal +gestational) (data not shown). In line with the triple-risk model and previous reports by us,[8,9] risk factors were divided into intrinsic and extrinsic categories (Fig 1). An intrinsic risk is defined as a genetic or environmental factor that affects susceptibility, including African American race, male gender, prematurity (<37 gestational weeks at birth), and prenatal maternal smoking or alcohol intake. An extrinsic risk is defined as a physical stressor around the time of death that may increase the risk of SIDS for an already vulnerable

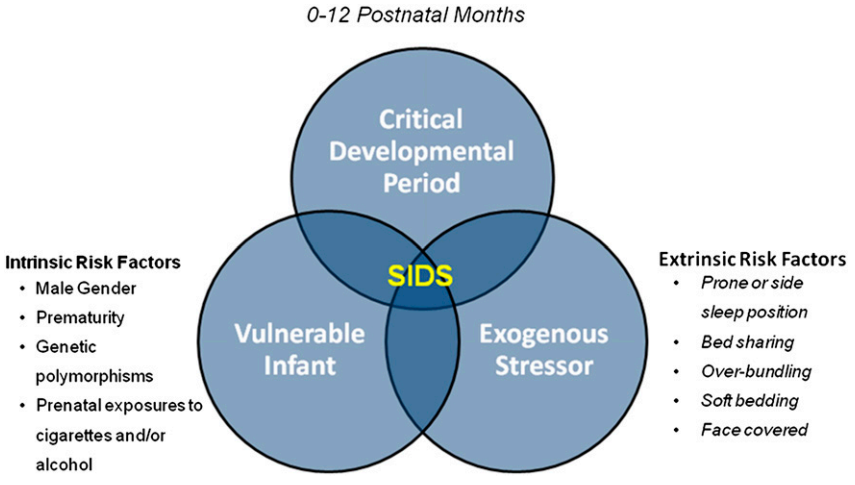

FIGURE 1
The triple-risk model for SIDS. Factors contributing to the vulnerability (bottom left circle) may include intrinsic risk factors. The exogenous stressors (bottom right circle) are the extrinsic risk factors for SIDS.

infant. These factors include being placed or found in a prone/side sleep position, found face-down, head covered, sleeping on an adult mattress, couch, or playpen, soft bedding, bed-sharing, and signs of upper respiratory tract infection (URTI). Although maternal smoking was considered to be an in utero (intrinsic) risk that may affect the infant's underlying vulnerability, it may also be considered a postnatal extrinsic risk, especially while bed-sharing.[10] A decision was made to avoid double-counting, based on literature suggesting that maternal smoking during pregnancy is the stronger risk.[11–13] In this regard, although the risk of African American race may reflect a particularly high burden of extrinsic risk factors, eg, bed-sharing,[14,15] in this population, we chose to place emphasis on the role of possible (intrinsic) genetic polymorphisms that may increase risk relative to race itself.[16,17] Information about every risk factor was not available in every case.

Continuous variables were summarized as mean ± SD and analyzed with *t* tests (2 groups) or analysis of variance (for >2 groups). Categorical variables were summarized as number (%) and analyzed with χ^2 tests. Categories with few cases were excluded from analysis, as was the category of "unknown." In all analyses, $P < .05$ was considered statistically significant. Analysis was performed by using SAS version 9.2 (SAS Institute Inc, Cary, NC).

RESULTS

SIDS cases ($N = 568$) accounted for 60% of all sudden unexpected infant deaths (Table 1). There was a sudden decrease in number of cases after the 1994 initiation of the BTS campaign, such that the SIDS rate decreased from 1.34 per 1000 births in 1991 to 0.64 per 1000 in 2008, with the decline observed across all races[18] (Fig 2). Although the average postnatal age of SIDS infants remained consistent over time (3.3 months), there was a trend away from the 2 to 4 months range in the BTS era, with slightly more younger and older infants (Table 2). The percentage of SIDS infants placed to sleep prone decreased from 85.4% to 30.1% ($P < .001$), whereas those placed supine increased from 1.9% to 41.7% ($P < .001$) and those found prone decreased from 84.0% to 48.5% ($P < .001$). The percentage of SIDS infants bed-sharing at the time of death increased from 19.2% to 37.9% ($P < .001$) (Table 2), especially among infants <2 months (29% vs 63.8%) (Table 3), and the percentage found in an adult bed increased from 23.4% to 45.4% ($P < .001$). The percentage born prematurely rose slightly from 20.0% to 29.0% ($P = .05$), whereas the percentage with a URTI decreased from 46.6% to 24.8% ($P < .001$).

Ninety-nine percent of SIDS infants had at least 1 intrinsic or extrinsic risk factor; 75% had at least 1 of each. The majority (57%) had at least 2 extrinsic risks and 1 intrinsic risk factor. The only significant association between SIDS risk factors was a negative association between bed-sharing and/or sleeping on adult mattresses with prone sleep position (infants bed-sharing or on adult mattresses were less likely to be sleeping prone; $P < .001$). There were 26 infants (4.6% of total; 5.5% of those with complete data) with no extrinsic risk factors and with known sleep position and bed-sharing status, 73% sleeping in a crib/stroller and 27% in a car/bouncy seat. Of 94 infants with only a single extrinsic risk factor, 51 were found prone, 14 side, and 29 with URTI. Forty infants were sleeping supine on an adult bed without documentation of face obstructed or soft bedding, 35 of them bed-sharing, without any additional extrinsic risk factors examined in this study. The ages and intrinsic risks of the infants with single or no extrinsic risk were similar to infants with multiple extrinsic risks. There was no significant change in the number of intrinsic or extrinsic risk factors over the study period (Table 2), with ~1.3 intrinsic (0–5 per infant) and 2.1 extrinsic (0–6 per infant) risks over time.

TABLE 1 SIDS and Other Sudden Unexpected Infant Deaths in San Diego County, California from 1991 to 2008

	SIDS, *n* (%)	Other Natural Causes, *n* (%)	Accident, *n* (%)	Homicide, *n* (%)	Unclassified, *n* (%)
1991–1993	169 (65)	38 (15)	27 (10)	9 (3)	17 (7)
1994–1995	92 (65)	20 (14)	11 (8)	11 (8)	8 (6)
1996–2008	307 (56)	73 (13)	75 (14)	35 (6)	62 (11)
Total	568 (60)	131 (14)	113 (12)	55 (6)	87 (9)

DISCUSSION

This study is the first long-term, hypothesis-driven analysis that addresses risk factors in SIDS infants in the United States before and after the initiation of the BTS campaign in a large cohort that has been rigorously characterized with standardized autopsies, postmortem ancillary studies, and death scene investigations. It is also the first to systematically examine in a large dataset the known SIDS risk factors subdivided into intrinsic and extrinsic categories, thereby uniting the findings within the context of the triple-risk model for SIDS. Our key finding is that, although the proportion of different risk factors for SIDS have changed relative to each other since the initiation of the BTS campaign, virtually all SIDS infants have at least 1 risk factor, and the majority (57%) have at least 1 intrinsic and 2 extrinsic risk factors. Moreover, the average number of risk factors per SIDS infant did not change with the initiation of the BTS campaign. Whereas prone sleep remains the most significant risk factor for SIDS, we found that sleeping on adult mattresses and bed-sharing, especially among younger

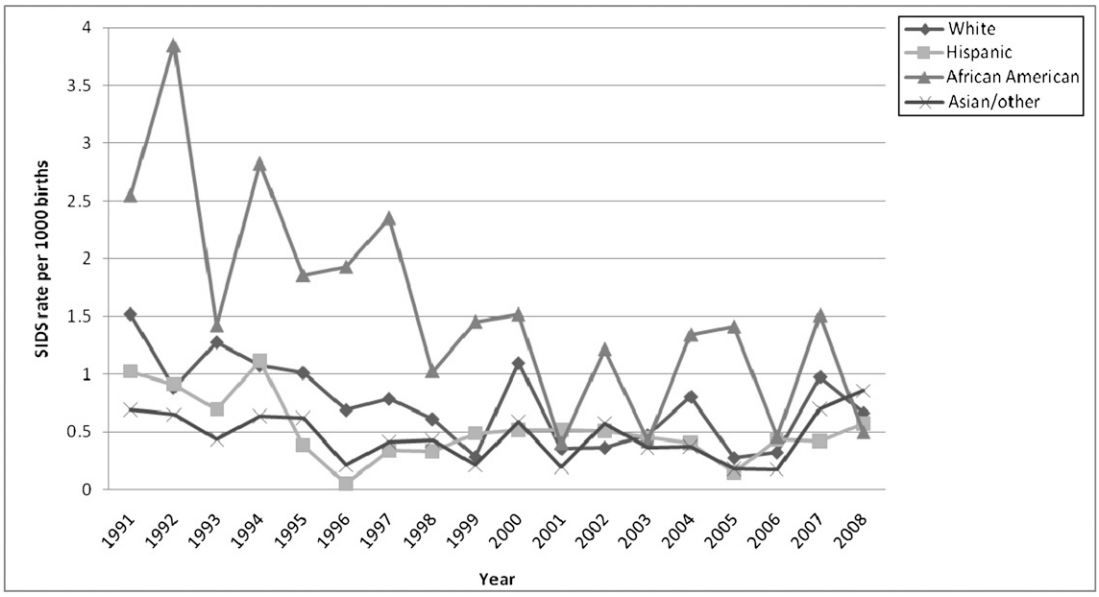

FIGURE 2

SIDS rate per 1000 births by year in San Diego County, California. The SIDS rate was calculated as the ratio of the number of SIDS deaths (San Diego Medical Examiner's Office) to the number of live births in San Diego County (California Department of Public Health Birth Records).[18] The BTS campaign in the United States began in 1994.

infants, have emerged as additional prominent risks. It is noteworthy that no evidence exists that more deaths are associated with bed-sharing in the BTS era, but rather that the higher proportion is due to declining rates of prone sleep-related deaths.

Despite the dramatic (65%) decrease in the incidence of infants placed prone to sleep after the initiation of the BTS campaign in San Diego County, still 30% of infants were reported to be placed prone. Moreover, the incidence of side placement, likewise a risky position, increased 133%, increasing to 27% even after the change in American Academy of Pediatrics guidelines in 2005. Clearly, enhanced efforts to educate caretakers about avoiding the prone and side sleep positions in infants are needed. Forty-two percent of infants placed side were found prone (44% of those <5 months), compared with only 25% of infants placed supine (19% of those <5 months). Taken together, these data suggest that some SIDS infants spontaneously roll to prone during the (unwitnessed) sleep period. An additional important point is that after the initiation of the BTS campaign almost one-third of the SIDS infants were found supine, underscoring the realization that SIDS will not be completely eradicated with the institution of universal supine infant sleep.

The occurrence of extrinsic risks in virtually all SIDS deaths implies that SIDS is precipitated by a "trigger" at the time of death. These extrinsic risk factors are consistent with asphyxia-generating conditions, eg, face-down position, prone position, and adult mattress. Research conducted in this same San Diego cohort over the same time frame suggests that infants may be vulnerable to SIDS because of a deficiency in the neurotransmitter serotonin in caudal brainstem regions that help mediate protective responses to homeostatic stressors such as asphyxia at the time of death.[8,9] Of critical importance, this study indicates that education about avoiding simultaneous and multiple risk factors, especially those that compromise oxygen exchange in the sleeping environment, is essential to help prevent SIDS, including among infants who may already be vulnerable.

Changes in SIDS Risk Profile Since the Initiation of the BTS Campaign

Consistent with previous studies, we identified a high prevalence of prone sleep, face-down or obstructed sleep, bed-sharing, maternal smoking, and URTI in SIDS infants across time periods. The high African American SIDS rate is consistent with past reports,[3] but the low absolute number of African American cases and the high proportion of Hispanics reflect the racial distribution of San Diego County. The current percentage of SIDS infants ever breastfed (78.7%) is lower than in the general population (92.0% in San Diego County in 2008).[19] Since initiation of the BTS campaign in 1994, the epidemiology of SIDS in the San Diego population has shifted away from the typical 2- to 4-month-old infant dying prone while having a minor illness. The rise in bed-sharing SIDS rates in this study is consistent with, although less dramatic than a United Kingdom study finding a rise from 12% to 50% from 1984 to 2003, also finding that the age of infants who bed-share was younger than before

TABLE 2 SIDS Risk Factors by Time Period[a]

	Pre-BTS 1991–1993	Transition Period 1994–1995	BTS Era 1996–2008	*P* value Pre Versus Post[b]
No. of SIDS cases	169	92	307	
Age, d[c]	100.2 ± 55.3, (91), 10–322	100.0 ± 56.6, (87.5), 20–276	101.3 ± 63.5, (86), 2–326	.85
Age[d]				.11
<2 mo	38 (22.5%)	22 (23.9%)	80 (26.1%)	
2–4 mo	111 (65.7%)	57 (62.0%)	173 (56.4%)	
≥5 mo	20 (11.8%)	13 (14.1%)	54 (17.6%)	
Gender				.58
Male	98 (58.0%)	61 (66.3%)	186 (60.6%)	
Female	71 (42.0%)	31 (33.7%)	121 (39.4%)	
Race				.14
White	83 (49.1%)	42 (45.7%)	135 (44.1%)	
African American	28 (16.6%)	16 (17.4%)	39 (12.8%)	
Hispanic	50 (29.6%)	28 (30.4%)	103 (33.7%)	
Other	8 (4.7%)	6 (6.5%)	29 (9.5%)	
Death in winter	69 (40.8%)	34 (37.0%)	138 (45.0%)	.39
Prematurity[e]	28 (20.0%)	30 (36.6%)	78 (29.0%)	.05
Twin or triplet[f]	9 (5.7%)	9 (10.2%)	16 (5.4%)	.90
Ever breastfed	52 (80.0%)	28 (84.9%)	155 (78.7%)	.82
Breastfed at time of death	26 (16.7%)	16 (18.4%)	73 (25.2%)	.04
Maternal smoking	37 (42.1%)	27 (37.0%)	78 (38.6%)	.58
Maternal alcohol use during pregnancy	7 (7.7%)	5 (6.8%)	8 (6.7%)	.77
Maternal drug use during pregnancy	18 (18.4%)	11 (14.5%)	15 (11.9%)	.18
Symptoms of URTI	75 (46.6%)	38 (44.2%)	72 (24.8%)	<.001
Sleep site[g]				<.001
Crib, bassinet, toddler bed, stroller lying flat	100 (59.9%)	41 (46.6%)	112 (36.6%)	
Adult bed	39 (23.4%)	37 (42.1%)	139 (45.4%)	
Couch, recliner, other soft chair[h]	12 (7.2%)	4 (4.6%)	18 (5.9%)	
Waterbed or inflatable bed	3 (1.8%)	1 (1.1%)	0 (0.0%)	
Car seat, car bed, bouncy chair	3 (1.8%)	0 (0.0%)	14 (4.6%)	
Floor, possibly on pillow or blanket	4 (2.4%)	2 (2.3%)	11 (3.6%)	
Playpen	3 (1.8%)	2 (2.3%)	10 (3.3%)	
Being held, breastfeeding, in a sling	3 (1.8%)	1 (1.1%)	1 (0.3%)	
Other	0 (0.0%)	0 (0.0%)	1 (0.3%)	
Unknown[i]	2	4	1	
Sleep position placed[j]				<.001
Prone	88 (85.4%)	32 (53.3%)	80 (30.1%)	
Side	10 (9.7%)	15 (25.0%)	60 (22.6%)	
Supine	2 (1.9%)	12 (20.0%)	111 (41.7%)	
Car seat/bouncy seat	2 (1.9%)	0 (0.0%)	14 (5.3%)	
Being held	1 (1.9%)	1 (1.7%)	1 (0.4%)	
Unknown[i]	66	32	41	
Sleep position found[j]				<.001
Prone	110 (84.0%)	49 (57.7%)	132 (48.5%)[k]	
Side	9 (6.9%)	18 (21.2%)	47 (17.3%)	
Supine	9 (6.9%)	18 (21.2%)	79 (29.0%)	
Car seat/bouncy seat	2 (1.5%)	0 (0.0%)	14 (5.2%)	
Being held	1 (0.8%)	0 (0.0%)	0 (0.0%)	
Unknown[i]	38	7	35	
Face position				.001
Down	47 (45.2%)	24 (34.3%)	78 (33.8%)	
Side	50 (48.1%)	32 (45.7%)	102 (44.2%)	
Up	7 (6.7%)	14 (20.0%)	51 (22.1%)	
Unknown[i]	65	22	76	
Face obstructed	12 (7.6%)	4 (4.6%)	16 (5.7%)	.44
Bed-sharing	32 (19.2%)	21 (25.0%)	114 (37.9%)[l]	<.001
Bed-sharing with other children	3 (1.8%)	2 (2.4%)	10 (3.4%)	.34
No. of known intrinsic risk factors[m]	1.3 ± 1.0, (1) 0–5	1.7 ± 1.2, (1) 0–5	1.4 ± 1.0, (1) 0–4	.70
≥1 intrinsic risk factor	134 (79.3%)	81 (88.0%)	260 (84.7%)	.14
≥2 intrinsic risk factors	66 (39.1%)	45 (48.9%)	110 (35.8%)	.49

TABLE 2 Continued

	Pre-BTS 1991–1993	Transition Period 1994–1995	BTS Era 1996–2008	*P* value Pre Versus Post[b]
No. of known extrinsic risk factors	2.1 ± 1.3, (2) 0–6	2.3 ± 1.2, (2) 0–5	2.1 ± 1.2, (2) 0–6	.78
≥1 extrinsic risk factor	155 (91.7%)	85 (92.4%)	274 (89.3%)	.39
≥2 extrinsic risk factors	109 (64.5%)	69 (75.0%)	211 (68.7%)	.35

[a] Continuous variables are summarized as mean ± SD, (median), range; categorical variables are presented as *n* (%). Some risk factors contain considerable missing data (eg, maternal smoking, alcohol, and drug use).

[b] Only the comparison between pre-BTS and the BTS era was tested, because 1994–1995 was considered a time of continual change in population practice. *t* test was used for continuous variables; χ^2 tests were used for categorical variables. The category "unknown" was always excluded from analysis.

[c] The interquartile range is 62–121 pre-BTS, 63.5–129.5 transition period, and 59–125 BTS era.

[d] Younger than 1 mo changed from 3.6% pre-BTS to 8.1% in the BTS era; 1 mo changed from 18.9% to 17.9%; 5 mo changed from 3.6% to 6.8%; 6+ months changed from 8.3% to 10.8%.

[e] Defined as <37 wk gestation.

[f] Thirty-three twins; 1 triplet post-BTS.

[g] Because of the inadequate sample size of most categories, the *P* value reflects a test of crib/bassinet/toddler bed/stroller lying flat versus adult bed. A toddler bed uses a crib mattress.

[h] Of these, almost all were sleeping on a couch: 8 pre-BTS, 4 transition period, and 17 BTS era. 47% of infants sleeping on a couch, recliner, or other soft chair were bed-sharing, remaining steady from 50% (*n* = 6) pre-BTS to 47% (*n* = 8) in the BTS era.

[i] The category "unknown" was excluded from statistical comparisons. It is presented only for completeness of data.

[j] Because of the inadequate sample size of some categories, the *P* value reflects a test of prone versus side versus supine.

[k] Fifty-nine percent prone from 1996 to 1999, 44% from 2000 to 2003, 30% from 2004 to 2008.

[l] Twenty-eight percent bed-sharing from 1996 to 1999, 44% from 2000 to 2003, 41% from 2004 to 2008.

[m] If male gender is excluded, female SIDS infants had an average of 0.75 to 1.07 other intrinsic risks across study periods.

their BTS campaign.[6] On the other hand, that study found an increase in the prevalence of maternal smoking from 57% to 86%; although we found a rate of ~40%, we did not see an increase, possibly because of lower smoking rates in the general US population. The average number of SIDS risks per infant has remained stable over time, supported by the negative association between bed-sharing and prone sleep, suggesting that those not sleeping prone were subject to alternative SIDS risks factors. The rise in proportion of prematurity among SIDS infants in the BTS era, consistent with a similar study in the United Kingdom,[6] is probably not due to an increase in premature births in the general population (10.0% in 1991 vs 11.2% in 2005 in California).[20] There was no SIDS risk profile unique to premature infants in the current study.

The small shift in age distribution toward a higher proportion of deaths in infants younger than 2 months and older than 4 months is consistent with previous findings[3,21–23] and explains the higher proportion of infants breastfed at time of death. Expanding literature suggests that bed-sharing is a risk for SIDS mostly among younger infants[15,24–30] and those whose mothers smoke.[10,15,24–27,31–34] These factors accounted for 63% of the bed-sharing cases in the current study, with a trend toward association between bed-sharing and smoking ($P = .13$). The basis of the slight increase in the number of older SIDS infants (>5 months) is currently unclear, although it is possible that some of these older infants are those in whom an underlying pathophysiologic process progresses over time, causing them to eventually succumb to SIDS even if they survive through the period of greatest risk. This possibility is in keeping with the finding of decreasing 5-hydroxytryptamine$_{1A}$ receptor binding with increasing postconceptional age in SIDS infants in certain regions of the medulla related to cardiorespiratory regulation, with the lowest levels in the oldest cases.[8,9] With the decline in prone sleep, infants also may not encounter an extrinsic SIDS risk until somewhat later in life. Older babies may be more likely to obstruct their face because of increased mobility and/or roll prone.

Influence of Intrinsic and Extrinsic Risk Factors in SIDS

As in our study, the concurrence of multiple risk factors and the rarity of risk-free SIDS[8,9,35–38] have been previously reported. A previous study[37] found at least 1 SIDS risk factor in 96% of cases; 78% had 2 to 7 risks, and only 2 (0.8%) cases were risk-free. Other studies emphasize the interaction between risk factors, with combinations of factors exhibiting more than additive odds of SIDS.[39–41] In the current study, there were no significant positive correlations between risk factors, suggesting that no specific risks cluster together, but rather any combination of risks raises the odds of SIDS. The majority of SIDS infants were subject to at least 2 extrinsic risk factors, suggesting that SIDS results from the simultaneous occurrence of multiple factors, rarely just one. This is likely why the proportion of SIDS infants with URTI dramatically decreased in the BTS era, because sick infants not sleeping prone are much less likely to succumb to SIDS. We found a trend toward association between URTI and prone sleep ($P = .08$).

On the basis of the triple-risk model (Fig 1), all SIDS infants are subject to an extrinsic risk factor, and they demonstrate, as well, an underlying vulnerability that may be associated in part with intrinsic risk factors, eg, male gender. The finding that ~90% of SIDS infants were subject to at least 1 known extrinsic risk supports the triple-risk

TABLE 3 SIDS Risk Factors by Age Group[a]

	Pre-BTS 1991–1993				BTS Era 1996–2008			
	<2 mo	2–4 mo	≥5 mo	*P* Value	<2 mo	2–4 mo	≥5 mo	*P* Value
No. of SIDS cases	38	111	20		80	173	54	
Twin or triplet	1 (2.7%)	7 (6.9%)	1 (5.3%)	.64	1 (1.3%)	14 (8.4%)	1 (2.0%)	.04
Ever breastfed	8 (72.7%)	41 (82.0%)	3 (75.0%)	.76	52 (88.1%)	80 (77.7%)	23 (65.7%)	.04
Breastfed at time of death	6 (16.7%)	18 (17.5%)	2 (11.8%)	.84	32 (41.0%)	32 (19.6%)	9 (18.4%)	<.001
Sleep site[b]				.48				<.001
Crib, toddler bed, stroller lying flat	19 (50.0%)	68 (62.4%)	13 (65.0%)		14 (17.7%)	80 (46.2%)	18 (33.3%)	
Adult bed	11 (29.0%)	24 (22.0%)	4 (20.0%)		50 (63.3%)	62 (35.8%)	27 (50.0%)	
Couch, recliner, other soft chair	5 (13.2%)	6 (5.5%)	1 (5.0%)		7 (8.9%)	9 (5.2%)	2 (3.7%)	
Waterbed or inflatable bed	0 (0.0%)	3 (2.8%)	0 (0.0%)		0 (0.0%)	0 (0.0%)	0 (0.0%)	
Car seat, car bed, bouncy chair	0 (0.0%)	2 (1.8%)	1 (5.0%)		4 (5.1%)	9 (5.2%)	1 (1.9%)	
Floor, possibly on pillow or blanket	1 (2.6%)	3 (2.8%)	0 (0.0%)		1 (1.3%)	8 (4.6%)	2 (3.7%)	
Playpen	1 (2.6%)	2 (1.8%)	0 (0.0%)		2 (2.5%)	5 (2.9%)	3 (5.6%)	
Being held, breastfeeding, in a sling	1 (2.6%)	1 (0.9%)	1 (5.0%)		1 (1.3%)	0 (0.0%)	0 (0.0%)	
Other	0 (0.0%)	0 (0.0%)	0 (0.0%)		0 (0.0%)	0 (0.0%)	1 (1.9%)	
Unknown[c]	0	2	0		1	0	0	
Sleep position found[d]				.66				.08
Prone	26 (81.3%)	74 (87.1%)	10 (71.4%)		23 (33.8%)	84 (53.9%)	25 (52.1%)	
Side	2 (6.3%)	6 (7.1%)	1 (7.1%)		14 (20.6%)	26 (16.7%)	7 (14.6%)	
Supine	3 (9.4%)	4 (4.7%)	2 (14.3%)		27 (39.7%)	37 (23.7%)	15 (31.3%)	
Car seat/bouncy seat	0 (0.0%)	1 (1.2%)	1 (7.1%)		4 (5.9%)	9 (5.8%)	1 (2.1%)	
Being held	1 (3.1%)	0 (0.0%)	0 (0.0%)		0 (0.0%)	0 (0.0%)	0 (0.0%)	
Unknown[c]	6	26	6		12	17	6	
Face position				.19				.03
Down	13 (56.5%)	25 (37.9%)	9 (60.0%)		12 (20.0%)	54 (39.7%)	12 (34.3%)	
Side	9 (39.1%)	37 (56.1%)	4 (26.7%)		35 (58.3%)	55 (40.5%)	12 (34.3%)	
Up	1 (4.4%)	4 (6.1%)	2 (13.3%)		13 (21.7%)	27 (19.9%)	11 (31.4%)	
Unknown[c]	15	45	5		20	37	19	
Face obstructed	2 (6.1%)	7 (6.5%)	3 (15.8%)	.35	1 (1.4%)	9 (5.6%)	6 (13.3%)	.02
Bed-sharing	11 (29.0%)	19 (17.4%)	2 (10.0%)	.16	51 (63.8%)	48 (28.7%)	15 (27.8%)	<.001
No. of known intrinsic risk factors	1.3 ± 1.0	1.3 ± 1.0	1.7 ± 1.2	.32	1.3 ± 0.9	1.5 ± 1.0	1.1 ± 0.9	.09
≥1 intrinsic risk factor	29 (76.3%)	87 (78.4%)	18 (90.0%)	.44	69 (86.3%)	149 (86.1%)	42 (77.8%)	.30
≥2 intrinsic risk factors	14 (36.8%)	43 (38.7%)	9 (45.0%)	.83	29 (36.3%)	65 (37.6%)	16 (29.6%)	.57
No. of known extrinsic risk factors	2.3 ± 1.1	2.1 ± 1.4	2.0 ± 1.4	.45	2.3 ± 1.2	2.0 ± 1.3	2.0 ± 1.3	.23
≥1 extrinsic risk factor	38 (100.0%)	100 (90.1%)	17 (85.0%)	.082	74 (92.5%)	152 (87.9%)	48 (88.9%)	.54
≥2 extrinsic risk factors	28 (73.7%)	69 (62.2%)	12 (60.0%)	.40	63 (78.8%)	114 (65.9%)	34 (63.0%)	.07

[a] Continuous variables are summarized as mean ± SD and analyzed with analysis of variance; categorical variables are presented as *n* (%) and analyzed with χ^2 tests. *P* values reflect differences between age groups within time period. Some risk factors contain missing data. The following risk factors showed no difference by age group and are therefore not included in the table: gender, race, death in winter, prematurity, maternal smoking, alcohol use, and drug use, URTI, and bed-sharing with other children.

[b] Because of the inadequate sample size of most categories, the *P* value reflects a test of crib/bassinet/toddler bed/stroller lying flat versus adult bed. A toddler bed uses a crib mattress.

[c] The category "unknown" was excluded from statistical comparisons. It is presented only for completeness of data.

[d] Because of the inadequate sample size of some categories, the *P* value reflects a test of prone versus side versus supine.

model, in which such risk factors represent 1 of the 3 interlocking circles of the Venn diagram. On the basis of complete data, 5% of the SIDS infants had no identifiable extrinsic risk factor. We speculate that this small subset of SIDS infants may be "excessively" vulnerable as a result of a particularly severe underlying pathophysiologic process, thereby requiring no extrinsic stressor to trigger the lethal event. This is a testable hypothesis for future studies in which a large sample size of SIDS infants without extrinsic factors is potentially available.

Potential Limitations of the Study

Potential limitations include the possibility of caregivers underreporting risks because of perceived stigma, lack of information concerning certain risk or protective factors, including pacifier use, socioeconomic status, timing/quantity/duration of smoke exposure, and parental alcohol and drug consumption before last sleep. Although there is no alternative to parental report for much of SIDS data collection, this study's findings were based upon standardized death scene investigation and autopsy reports. Data concerning risk factors in non-SIDS controls were unavailable for reference and estimation of odds ratios; thus, we are only able to evaluate which SIDS risk factors are most common, not which raise the odds of SIDS most significantly. Because data were not available for each case, the number of risks is probably

underestimated. Additional studies in other geographical areas are warranted to determine if findings from this San Diego cohort can be generalized to SIDS throughout the country. It is conceivable that the large Hispanic population of San Diego County might be associated with culturally distinct infant care practices that somewhat alter the SIDS risk factor profile, although similarities to previously reported populations were observed. Given the prevalence of multiple testing, *P* values close to .05 (eg, breastfeeding in Table 3) should be interpreted cautiously and require confirmation in other studies.

CONCLUSIONS

The profile of SIDS risk factors changed in San Diego County after the initiation of the BTS campaign. The number of risk factors per case, however, remained steady over time, with most infants subject to at least 1 intrinsic risk factor and at least 2 extrinsic risks. Although SIDS is a disease,[8,9] and these risk factors are not causative of SIDS in and of themselves, risk reduction campaigns that emphasize the importance of avoiding multiple and simultaneous SIDS risks are essential. Although prone sleep alone is a large risk factor for SIDS, there is evidence that some risks generally appear in conjunction with other factors. Thus, if caretakers are unable to meet all ideal sleep conditions, this study suggests that meeting as many as possible will still be beneficial.

ACKNOWLEDGMENTS

The authors thank the deputy medical examiners and scene investigators of the Office of the Medical Examiner of San Diego County, California for their continuous support. The authors also thank Dr Lynn A. Sleeper, ScD (New England Research Institutes) and Dr Eugene Nattie, MD for helpful comments during manuscript preparation.

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Pediatrics originally published online March 26, 2012;

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