

# Concurrent Risks in Sudden Infant Death Syndrome

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## KEY WORDS

sudden infant death syndrome, SIDS risk factors, maternal smoking, prone sleep

## ABBREVIATIONS

SIDS—sudden infant death syndrome

AAP—American Academy of Pediatrics

SCNJ—SIDS Center of New Jersey

URI—upper respiratory infection

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**WHAT'S KNOWN ON THIS SUBJECT:** The incidence of SIDS has declined since the onset of risk-reduction education, although it remains the leading cause of infant mortality from 1 month to 1 year of age. The inadequate dissemination of risk-reduction strategies has impeded progress.



**WHAT THIS STUDY ADDS:** Our study results raise awareness of the rarity of SIDS without risk factors, the predominance of multiple risks, the patterns of co-occurrence, and the associations between modifiable and nonmodifiable risks, leading to recommendations for more inclusive and comprehensive risk-reduction education.

## abstract

**BACKGROUND:** Despite improved education on safe sleep, infants are still exposed to multiple risks for sudden infant death syndrome (SIDS). Variability among health care providers continues to exist regarding knowledge of risk factors and the provision of education to caregivers.

**OBJECTIVE:** To enhance the content and delivery of SIDS risk-reduction initiatives by physicians and other health care providers and to provide them with a context for evaluating their discussions of risks and compensatory strategies, we sought to raise awareness of the frequency of risk factors in SIDS cases, patterns of co-occurrence, associations between modifiable and nonmodifiable risks, and the rarity of cases without risk.

**DESIGN AND METHODS:** In a population-based retrospective review of 244 (97%) New Jersey SIDS cases (1996–2000), we assessed the frequencies and co-occurrences of modifiable (maternal and paternal smoking, nonsupine sleep or prone status at discovery, bed-sharing, or scene risks) and nonmodifiable (upper respiratory infection or <37 weeks' gestational age) risks.

**RESULTS:** Nonsupine sleep occurred in 70.4% of cases with data on position (159 of 226). Thirteen cases were of infants who were discovered prone, with an increased positional risk to 76.1%, in which 87% contained additional risks. Maternal smoking occurred in 42.6% (92 of 216) of the cases with data on this risk, and 98% among those cases had additional risks. At least 1 risk was found in 96% of the cases, and 78% had 2 to 7 risks. Of the 9 of 244 risk-free cases (3.7%), 7 lacked data on 2 to 5 risks per case. On the basis of the complete data, only 2 (0.8%) of all 244 cases were risk free. When nonmodifiable risks were excluded, 5.3% of the cases met this definition.

**CONCLUSIONS:** Risk-free and single-risk SIDS cases are rare, and most contain multiple risks. Parent education should be comprehensive and address compensatory strategies for nonmodifiable risks. *Pediatrics* 2010;125:447–453

Despite improved education on safe sleep practices,<sup>1–5</sup> living infants are still exposed to multiple risks that are associated with sudden infant death syndrome (SIDS).<sup>6–9</sup> Variability among health care providers continues to be found with respect to the knowledge of risks and the provision of education to parents and other caregivers.<sup>10–13</sup>

Growing demands for preventive health counseling during office visits,<sup>14</sup> divergent personal beliefs regarding safety and efficacy,<sup>11</sup> disparity in the translation of SIDS guidelines across health care professions that provide risk-reduction education,<sup>10,13</sup> or controversy, as is the case with bed-sharing,<sup>15</sup> may be some of the factors that impinge on adequate dissemination of the SIDS risk-reduction guidelines of the American Academy of Pediatrics (AAP),<sup>16</sup> specifically with respect to the range and consistency of the information. Conflicting or inadequately presented guidance can have the unintended consequence of promoting or, at best, failing to diminish the barriers to compliance.<sup>17–19</sup>

To promote comprehensive education of parents and other caregivers on methods for reducing the risk of SIDS, and to give physicians and other health care providers a context for reviewing the thoroughness of their educational initiatives, we sought to document the frequency and co-occurrence of risks in SIDS cases and to characterize the combinations of risk. We hypothesized that multiple risks represent the most common pattern of occurrence in SIDS and that SIDS without risk is rare. We conducted a large population-based review of SIDS cases to describe the frequency of each risk, the quantity of risks per case, the presence of SIDS without risk, and the frequency and characteristics of co-occurrence patterns, including those with known interaction effects.<sup>20–22</sup> Previous studies that had examined multiple risks<sup>23,24</sup>

did not quantify risk-free status or risk frequency, except when limited to 2 risks<sup>21,25</sup> or in studies limited by sample size and the absence of data on smoke exposure.<sup>26–29</sup>

## METHODOLOGY

### SIDS Cases

Under a grant from the New Jersey Department of Health and Senior Services, the SIDS Center of New Jersey (SCNJ) receives data on all cases of sudden infant death finalized as SIDS in New Jersey. For our descriptive study, we conducted a population-based, retrospective database review of all deaths that were coded as being caused by SIDS from 1996 through 2000.

In New Jersey, a diagnosis of SIDS is made via a medical-examiner system that includes an autopsy and a death-scene investigation. Final coding is provided by the New Jersey Center for Health Statistics. For all of the cases with available data, we determined the percentage of cases with modifiable risks (nonsupine placement at last sleep,<sup>16</sup> maternal and paternal smoking [prenatal or postnatal status not specified],<sup>30,31</sup> bed-sharing with an adult,<sup>16</sup> and scene risks [the use of sofas, quilts, blankets, pillows, or the presence of other children])<sup>16,24</sup> and with 2 risks that are not directly modifiable (prematurity [born at <37 weeks' gestational age<sup>32</sup>] and upper respiratory infection<sup>22</sup> [URI]) as reported by the caregiver and/or medical examination. Although our data did not specify whether smoking occurred during or after pregnancy, each period added risk.<sup>31</sup> Paternal smoking also served as an independent contributor.<sup>30</sup> We also determined the number and type of concurrent risks and the number of cases with risk-free status. The number of cases without risk was first calculated on the basis of all risks and then limited to those risks that

were defined as modifiable. Where noted, prone status at death was also included as a risk for cases in which it represented a shift from supine placement. The chronological and corrected ages of those infants who shifted from supine to prone positions and those who remained supine were calculated. We did not further characterize the details of the nonsupine-sleep risk in terms of lateral or prone status or of scene risks, because these data have been reported elsewhere.<sup>33</sup> For each ratio presented below in which the frequency of a risk is the numerator, we further describe the basis of the denominator as 1 of the following: all cases ( $N = 244$ ), wherein missing data were conservatively defined as the absence of risk, or all cases in which the risk factor under examination was reported as positive or negative (hence, all cases that were free of missing data). When 2 risks are combined into a larger concept, such as with the conversion of maternal and paternal smoking status into a variable that documented smoke exposure by either or both parents, the denominator represents all cases in which a positive finding was reported in at least 1 parent and in which a negative finding was based on a documented negative finding rather than on missing data. Race was based on self-report. No income data were available. Data were deidentified. This study received institutional review board approval as meeting the standards for exempt status.

### Statistical Methods

Groups were compared by using  $\chi^2$  tests for categorical data and  $t$  tests for independent samples with continuous data. Two-tailed probabilities were used.  $P < .05$  defined statistical significance. Analyses were completed by using Statistica 5.5 (StatSoft Inc, Tulsa, OK).

**TABLE 1** Demographic Characteristics of SIDS Cases

| Variable                | Cases With Reported Data, <i>n</i> |                            |
|-------------------------|------------------------------------|----------------------------|
| Black race <sup>a</sup> | 244                                | 121 (49.6) <sup>b</sup>    |
| Gestational age, wk     | 228                                | 37.62 (3.67) <sup>c</sup>  |
| Age at death, d         | 243                                | 94.32 (38.07) <sup>c</sup> |
| Prenatal care           | 180                                | 162 (90) <sup>b</sup>      |
| Maternal age, y         | 212                                | 24.67 (6.17) <sup>c</sup>  |
| Gravida > 2             | 210                                | 95 (45.24) <sup>b</sup>    |
| Breastfeeding           | 184                                | 52 (28.26) <sup>b</sup>    |

<sup>a</sup> Race classification was based on self-report.

<sup>b</sup> Data are reported as *n* (%).

<sup>c</sup> Data are reported as mean (SD).

## RESULTS

### Individual Risks in SIDS Cases

From 1996 through 2000, 244 of the 251 SIDS cases that occurred in New Jersey (97%) were reported to the SCNJ. Demographic data are presented in Table 1. The number and percentage of cases with each study-defined risk is shown in Table 2. Each risk in Table 2 was calculated as a proportion of the cases for which the presence or absence of the named risk was reported. If each risk was calculated as a ratio of all 244 cases, the percentage of positive findings for each risk was slightly lower and ranged from 70.5% for the position risk (nonsupine sleep place-

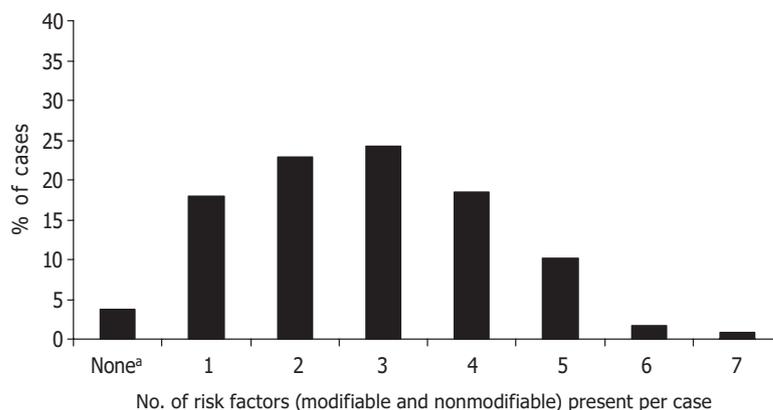
**TABLE 2** Risk Factors in SIDS Cases

| Variable                                  | <i>N</i> <sup>a</sup> | <i>n</i> (%) |
|---|-----------------------|--------------|
| Position                                  |                       |              |
| Nonsupine sleep placement                 | 226                   | 159 (70.4)   |
| Nonsupine placement or discovery prone    | 226                   | 172 (76.1)   |
| Smoking                                   |                       |              |
| Maternal smoking                          | 216                   | 92 (42.6)    |
| Paternal smoking                          | 180                   | 90 (50.0)    |
| Smoking by 1 or both parents <sup>b</sup> | 204                   | 123 (60.3)   |
| URI                                       | 209                   | 92 (44.0)    |
| Scene risks <sup>c</sup>                  | 238                   | 75 (31.5)    |
| <37 wk gestational age                    | 228                   | 62 (27.2)    |
| Bed-sharing                               | 239                   | 93 (38.9)    |

<sup>a</sup> *N* indicates the number of the 244 study cases for which data were reported on the variable indicated.

<sup>b</sup> Cases with a positive risk for 1 parent or negative risk status for both parents based on negative reports rather than missing data.

<sup>c</sup> Scene risks include the use of quilts, blankets, pillows, or sofa or the presence of other children.

**FIGURE 1**

The number of risk factors per SIDS case. Risk factors were defined as nonsupine placement (including 13 infants who were placed in a supine position but discovered prone), maternal smoking, URI, scene risks, paternal smoking, birth <37 weeks' gestational age, and bed-sharing. Of the 9 of 244 cases (3.7%) with no reported study-defined risks, 7 were missing data on 2 to 5 risks per case, precluding a definitive exclusion. On the basis of complete data, 2 cases (0.8% of all cases) were risk free.

ment or discovery prone after supine placement) to 25.4% for prematurity.

### Concurrent Risks

One to 7 risks were reported as being present in 96.3% of all 244 cases (Fig 1). The percentage of cases with multiple concurrent risks ranging from 2 to 7 was 78.3% (191 cases). A single risk was reported in 18% (44 cases). Specifically, single-risk status was noted in 12.8% (22 of 172) of cases of infants who slept in a nonsupine position or who were prone at discovery, 2.2% (2 of 92) of maternal-smoking cases, 10% (9 of 92) of URI cases, 1.1% (1 of 90) of paternal-smoking cases, 8.1% (5 of 62) of cases with infants who were <37 weeks' gestational age, and 5.4% (5 of 93) of bed-sharing cases. All scene-risk cases contained other risks. In the remainder of the cases exposed to each risk, other risks were also present. Table 3 describes these co-occurrence patterns in the cases that were positive for each risk.

### Risk-Free SIDS Cases

Nine of the 244 cases (3.7%) had no positive risk factors. However, 7 were missing data on 2 to 5 risks per case, precluding a definitive exclusion of

risk. Thus, on the basis of complete data, only 2 cases (0.8% of all 244) were free of risk.

Of the 44 of 244 cases that reported a single positive risk factor, 9 cited URI and 5 cited prematurity as the exclusive risks. In addition, 2 of the 56 cases for which 2 risks were reported were based on the combination of prematurity and gestational age. If we were to limit our calculation of risk to those that were defined as modifiable, these 16 cases with risk that was limited to the presence of prematurity and/or URI would be risk free, thereby increasing such cases from 9 to 25 of 244 (10.2%). However, 4 of the 9 cases with URI and 1 of the 5 with prematurity were missing complete data regarding 1 or more of their remaining risks. Thus, the cases for which risk-free status could be definitively established would rise by 11 rather than 16, increasing from 2 to 13 (5.3%).

### Age at Death and Position Shift

Infants who were placed in a supine position but were discovered prone were not significantly older than those who remained supine in terms of chronological age ( $120.3 \pm 60.8$  vs  $96.4 \pm$

**TABLE 3** Co-occurrence of Risks

| Additional Risk        | Type of Risk Present, % (n/M) With Additional Risks <sup>a,b</sup> |                |              |              |                |                        |              |
|------------------------|--|----------------|--------------|--------------|----------------|------------------------|--------------|
|                        | Nonsupine <sup>c</sup>   | Mother Smoking | URI          | Scene        | Father Smoking | <37 wk Gestational Age | Bed-Sharing  |
| Nonsupine              | —  | 76.4 (68/89)   | 72.3 (60/83) | 83.6 (61/73) | 74.7 (65/87)   | 69.5 (41/59)           | 80.0 (68/85) |
| Mother smoking         | 44.2 (68/154)  | —              | 40.2 (33/82) | 53.6 (37/69) | 69.4 (59/85)   | 54.6 (30/55)           | 53.1 (43/81) |
| URI                    | 40.8 (60/147)  | 39.3 (33/84)   | —            | 28.8 (19/66) | 41.0 (34/83)   | 40.4 (23/57)           | 45.0 (36/80) |
| Scene                  | 36.1 (61/169)  | 40.2 (37/92)   | 20.9 (19/91) | —            | 37.1 (33/89)   | 28.3 (17/60)           | 43.0 (40/93) |
| Father smoking         | 50.4 (65/129)  | 85.5 (59/69)   | 47.2 (34/72) | 58.9 (33/56) | —              | 59.1 (26/44)           | 54.6 (36/66) |
| <37 wk gestational age | 24.8 (41/165)  | 33.3 (30/90)   | 25.8 (23/89) | 24.6 (17/69) | 30.2 (26/86)   | —                      | 30.6 (26/85) |
| Bed sharing            | 39.8 (68/171)  | 46.7 (43/92)   | 40.0 (36/90) | 53.3 (40/75) | 40.0 (36/90)   | 42.6 (26/61)           | —            |

<sup>a</sup> Cases positive for the additional risk/all cases positive for the risk designated in each column.

<sup>b</sup> Denominators represent all cases with complete data on the status of the additional risks named in the first column.

<sup>c</sup> In addition to the cases in which the infants were placed to sleep in a nonsupine position, this category includes those infants who were placed supine but found prone.

58.1 days;  $P = .19$ ) or of age adjusted for gestation length ( $110.5 \pm 58.7$  vs  $77.2 \pm 66.3$  days;  $P = .11$ ). When we adjusted for gestational age, the age range for those infants who shifted sleep positions ranged from <1 to 7.7 months compared with <1 to 11 months for those who did not.

## DISCUSSION

### Overview

In a retrospective descriptive study of a 5-year population of SIDS cases, we reviewed the presence of the risk factors of nonsupine sleep or a shift from supine to prone sleep at discovery, maternal and paternal smoking, bed-sharing, scene risks (such as the presence of other children or blanket or sofa use) prematurity, and URI. We found that (1) SIDS in the absence of risk was rare, even when risk factors were limited to those described as being modifiable, (2) multiple concurrent risks characterized the majority of cases, and (3) nonmodifiable risks were commonly accompanied by modifiable ones.

These findings support the importance of having comprehensive risk-reduction education, with attention to multiple risks, and of helping families with nonmodifiable risks to adopt risk-lowering strategies. These data may also help providers offer more-detailed characterizations of the presence of risk.

### Relationship of Findings to Previous Studies

Other studies that have identified cases that were free of risk have found, as we did, that SIDS without risk is rare.<sup>26–29</sup> However, our study contains at least twice as many cases and provides data on smoking as well as patterns of co-occurrence. In a study of cases from 1992 to 1997, 98% of the 115 cases with available data were of infants who were found in a prone position, sleeping outside of a crib, or sleeping with another person.<sup>26</sup> Eighty percent of our cases also contained 1 or more of these 3 characteristics on the basis of all cases with either a positive report for at least 1 risk or a negative report for all 3. In a more recent (2001–2004) database of sudden unexpected infant deaths, the final causes of which included undetermined and position-related asphyxia in addition to SIDS, 15% were free of 1 or more potential risk factors for asphyxia.<sup>29</sup> However, 68% of these infants had other risks including URI and prematurity, leaving only 5% of the infants with no potential risks for asphyxia, illness, or other pertinent medical history. In this study by Pasquale-Styles et al,<sup>29</sup> 67 cases (32%) were classified as SIDS. Finally, authors of a study of 102 cases reported that 7.8% of the infants slept alone, in a crib or bassinet, and on their back or side,<sup>27</sup> and 90% of 31 SIDS cases of infants who were studied for

brainstem abnormalities were exposed to at least 1 stressor at death, with 77% of the infants having either or both nonsupine-sleep and bed-sharing risks.<sup>28</sup>

### Risk-Reduction Education

The triple-risk model of SIDS characterizes the risk of an adverse outcome as resulting from the confluence of a physiologically vulnerable infant challenged by an environmental stressor in a developmental period that is not yet supported by compensatory mechanisms.<sup>34</sup> Until it is possible to detect and correct relevant vulnerabilities of living infants, comprehensive risk reduction that represents all of the available guidance remains the methodology that is most often associated with reduced occurrences of SIDS.<sup>16,28</sup>

The combination of modifiable and nonmodifiable risk factors has implications for caregiver education. In New Jersey, 9% of the births during our study period were preterm,<sup>35</sup> compared with 27% of our cases. The rate of preterm births is increasing,<sup>36</sup> as is the need to help families learn about compensatory strategies for this nonmodifiable risk factor.<sup>16</sup> Preterm birth further elevates the increased risk for SIDS that is already posed by prone and side sleep.<sup>21,25</sup> The reduction in spontaneous arousals<sup>37</sup> and the slowing electrocortical activity<sup>38</sup> in prone-sleeping preterm infants provide in-

sight into the underlying mechanisms of SIDS. Yet, very low birth weight infants are more likely to be placed in a prone position,<sup>39</sup> perhaps in response to what parents observe or are told during hospitalization.<sup>16</sup> In our study, two thirds of the preterm infants had been placed to sleep in a nonsupine position. The AAP guidelines advise medical providers to help families understand and adopt the transition from therapeutic prone positioning in the NICU to supine sleep after discharge.<sup>16</sup> However, discharge instructions may not consistently reflect this recommendation.<sup>11</sup> Educational interventions in both newborn care units and NICUs have been developed for nurses.<sup>5,13</sup> Health care providers can work with hospital-based nurse educators to implement these programs. After discharge, physicians must facilitate the continued use of supine placement, along with other risk-reduction guidance, by discussing safe sleep with parents at each outpatient visit. Reinforcement of the message may also counteract the decline in compliance over the first year of life.<sup>8</sup>

The second nonmodifiable risk, URI, was found in 44% of the study cases. Reduced arousal in quiet sleep with infection suggests it is a mechanism for elevated risk.<sup>40</sup> The combination of a recent illness or infection with prone sleeping increases the risk of SIDS over the level found for the individual risk factors.<sup>20,22</sup> Approximately three quarters of the infants in our URI cases were placed in a nonsupine position. In addition to decreasing the potential impact of infection, supine sleep has been associated with less fever at 1 month of age, fewer stuffy noses at 6 months, and a decline in outpatient visits for ear infections at 3 and 6 months.<sup>41</sup> Smoking has been shown to elevate the risk that infectious symptoms present.<sup>22</sup> We found that 40% of mothers and 47% of fathers smoked in

cases with URI. As with other risk factors, exposure to tobacco smoke reduces arousal to hypoxic challenges.<sup>42</sup> Winickoff et al found that pediatricians did not ask enough questions or provide sufficient counseling on smoke exposure or assist families with access to smoking-cessation interventions.<sup>10</sup> Moreover, given the greater presence of household smoking in black families with infants, relative to white and Hispanic families,<sup>7</sup> and the disparity in SIDS rates among these groups,<sup>16</sup> we further recommend that health care providers work to reduce the disparity in access to smoking-cessation interventions.<sup>42</sup>

Finally, breastfeeding has been associated with reduced infection.<sup>43,44</sup> However, the 2005 updates to the SIDS risk-reduction guidelines noted that there was insufficient evidence to include breastfeeding as a risk-reduction strategy. More recently, Vennemann et al<sup>45</sup> found that breastfeeding reduced the risk of SIDS by 50% and recommended that advice on breastfeeding be added to SIDS risk-reduction guidelines.

### Addressing Limitations

We were not concerned with time-related recall bias in this study because of the immediacy of postmortem data-gathering. Even in a study with a median lapse of 3.8 months between an infant's death and the subsequent interview,<sup>46</sup> the coroners' records and maternal reports reached moderate agreement. Because our study design did not include comparisons with living infants, there were no issues concerning relative recall bias between these groups.

However, we must consider the possibility of the underreporting of risk because of a perception of stigma.<sup>47</sup> For example, a study that compared the reported position at discovery to a potentially corroborating physical sign noted contradictory findings that sug-

gested the prone position was underreported at death.<sup>48</sup> We cannot rule out that concerns about stigma may have accounted for the reports of supine placement in some of the very young infants who were found prone. However, even with potential bias and despite missing data, risk was reported in nearly all cases.

Our data set covers the period from 1996 to 2000. Given the prospective time required to prepare sufficiently large data sets on cases for SIDS research, it is not uncommon for studies published after the 2000 or 2005 revisions to the AAP guidelines on SIDS risk reduction to use data sets from earlier periods.<sup>49</sup> Because of the low annual rate of SIDS in New Jersey, we faced a challenge in preparing large, era-specific databases. However, before using these data, we concluded that they could provide useful findings regarding risk despite the evolution in the guidelines. To support this conclusion, we noted that the variables we examined in this study had been identified as risk factors in the past<sup>20,23</sup> and currently exist. It is interesting to note that in 1996 and 1997, the AAP Task Force on Infant Positioning and SIDS emphasized supine sleep as conveying the lowest risk, addressed the avoidance of soft surfaces in the sleep environment, suggested the inclusion of preterm infants in its positional recommendation, and addressed the potential risks of a bed-sharing environment.<sup>50,51</sup> Moreover, the SCNJ presented these recommendations in educational programs throughout New Jersey during the era addressed in the database.<sup>52</sup> Although the rate of SIDS has declined, risk factors continue to exist for living infants,<sup>7,8</sup> and interactions between the risk factors recorded in Pregnancy Risk Assessment Monitoring System data from 2006 parallel those in our database.<sup>9</sup> For example, our cases showed a disproportional

tionate presence of maternal smoking in premature infants compared with term infants (55% vs 38%;  $P = .03$ ), a finding that is also reflected in the Pregnancy Risk Assessment Monitoring System database on living infants, albeit in smaller percentages.

The mean age at death in the cases of infants who were reported to have shifted from supine sleep to the prone position on discovery was younger than the age at which supine-sleeping infants achieve this milestone.<sup>53,54</sup> Thus, the mechanisms for at least a portion of the shifts in cases reported in our study remain unknown.

Another limitation is that our study was descriptive and not a case-control

study. Finally, we urge caution in generalizing these findings to communities that are demographically dissimilar to New Jersey.

## CONCLUSIONS

A review of SIDS cases revealed that multiple risk factors were prevalent. Most of the cases contained the presence of more than 1 risk. Nonmodifiable risks occurred with modifiable risk factors in patterns that have been found elsewhere to further elevate risk. Risk-free status was rare. These findings underscore the importance of providing comprehensive risk-reduction education to parents and other caregivers. Future studies are needed to assess trends in the pat-

terns of risk in SIDS cases as the AAP guidelines, risk-reduction campaigns, death-scene–investigation techniques, and diagnostic criteria evolve. Case-control studies are also needed to assess the relative risk not only of single risks but also of combinations of risk.

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