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Sudden Infant Death Syndrome Risk Factors with Regards to Sleep Position, Sleep Surface, and Co-Sleeping*

ABSTRACT: We present a study of 102 Sudden Infant Death Syndrome (SIDS) deaths using a retrospective review of medical examiner autopsy reports. The prevalence of sleep related risk factors with regards to sleep surface, sleep position, and co-sleeping were determined in a population of infants less than 1-year-old. Of the 102 SIDS deaths, 67 (65.7%) were not in a crib, 63 (61.8%) were prone, and 48 (47.1%) were co-sleeping. However, 94 (92.2%) of these deaths had at least one risk factor present. Only 8 (7.8%) infants had slept alone, in a crib or bassinet, and on their back or side. Infants less than 4-months-old had a higher rate of co-sleeping (54.7%) than the older infants (25.9%), and a higher frequency of heart malformations at post-mortem examination. The older infants were more likely to exhibit pulmonary and tracheal inflammation, and neuropathology.

KEYWORDS: forensic science, sudden infant death syndrome, risk factors, co-sleeping, prone position

Sudden Infant Death syndrome (SIDS), defined as "the sudden death of an infant less than 1 year of age which remains unexplained after a thorough case investigation, including performance of a complete autopsy, examination of the death scene, and review of the clinical history," (1) is a leading cause of infant death beyond the neonatal period within the United States (2). SIDS is most common in infants 1–4 months of age, with a peak incidence at 2 months (3,4). Although the etiology of SIDS largely remains unknown, abnormalities in fatty acid oxidation (5–7), organic acidemia (7), and prolongation of the QT interval predispose some infants to SIDS (8).

A well documented risk factor for SIDS is the prone sleep position (9–19). The importance of prone sleeping has been emphasized by the success of the "back to sleep" campaign within the United States. The American Academy of Pediatrics first recommended a supine sleeping position for infants in 1992, and the "back to sleep campaign" was initiated in 1994. The campaign resulted in a decrease in the frequency of infant prone sleeping from >70% to ~20% with a concomitant drop in the SIDS rate by >40% (4). Reductions in SIDS have also been seen in other countries that have undertaken programs to reduce prone sleeping (15,20–22).

Other suggested risk factors for SIDS include co-sleeping (23,24), sleeping outside of a crib or bassinette (25–27), side sleeping position (23), unsafe bedding (14), exposure to tobacco smoke (28–30), parental drug use, low birth weight/prematurity (31,32), young maternal age (33), infection (34), and poor socioeconomic status (35). Often a combination of risk factors is present (11–13,16,17,19,36,37).

Adult beds, couches, sofas, and waterbeds are independent risk factors for infant death due to the potential for entrapment, wedging, and smothering (25,27,38–40). Use of these sleep surfaces may be associated with co-sleeping and the risk of asphyxia due to overlaying (16,17,26,27,36,40). Although it is well accepted that co-

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sleeping with an adult smoker is a significant risk for SIDS, the risk that a non-smoking adult poses is controversial (11,23,24,41–43).

The current study is a retrospective review of all infant deaths in North Carolina during 1999 and 2000 that could possibly be considered SIDS. Similar to Kemp et al. (2000), the current study was not undertaken to determine the risk factors for SIDS. Rather, we report the prevalence of already established risk factors with regards to sleep position, sleep surface, and co-sleeping. The overlap of these risk factors in the study population is reported. Of particular interest was the frequency of infants who died with no risk factors present.

Methods

The North Carolina Office of the Chief Medical Examiner's records were retrospectively reviewed for all infants aged 0–1 year who died suddenly and unexpectedly during 1999 and 2000. Infants who had causes of death such as accidental asphyxia or aspiration were included in the study due to the possibility of diagnostic overlap with SIDS. Excluded were infants with a well established history of natural disease that led to the death of the infant. Also excluded were deaths due to motor vehicle accident or other trauma.

The study was further limited to infant deaths in which the sleep surface, sleep position, and occurrence of co-sleeping were known. These data were collected from autopsy and scene investigation reports. If an infant died when co-sleeping, the number and identity of all co-sleepers was determined. The cause of death, manner of death, and anatomic diagnoses on the autopsy reports were tabulated for all of the infant deaths. The data were analyzed in the following three populations: all infants, infants up to 4-months-old, and infants more than 4-months-old. Four months was chosen as a cutoff due to the rapid decline in SIDS rates in infants older than this (3,4).

Results

The study population included 102 infant deaths. Seventy-five (73.5%) were younger than 4-months-old, and 27 (26.5%) were older than this. The gender of the infants was 57 (55.9%) male and

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45 (44.1%) female. The ethnicity of the infants was 53 (52.0%) white, 46 (45.1%) black, 1 (1.0%) Hispanic, and 2 (2.0%) other. The ages ranged from 3 to 338 days with average and median ages of 96 and 78 days, respectively. The causes of death were reported as SIDS in 80 (78.4%), asphyxia in 10 (9.8%), aspiration in 1 (1.0%), and undetermined in 11 (10.8%). The manner of death was determined to be natural in 81 (79.4%), accidental in 11 (10.8%), and undetermined in 10 (9.8%).

The most common sleep surface on which an infant was found was an adult mattress, with 37 (36.3%) infants found in this location (Table 1). Only 35 (34.3%) infants were found in a crib or bassinet. Thus, 67 (65.7%) of the infants were found not sleeping in a crib or bassinet. Infants younger than 4-months-old were slightly more likely to be found sleeping outside of a crib or bassinet (68%) than older infants (59.3%). Fourteen (13.7%) infants were found on couches or sofas, and 2 (2.0%) were found on water beds. Other sleep surfaces on which infants were found included floors, chairs, car-seats, play-pens, and piles of clothes.

Prone was the most common sleep position in which the infant was found, and this position accounted for 63 (61.8%) deaths (Table 2). The rate of prone sleeping was similar for both infants younger than 4-months-old (61.3%), as well as older than this (63.0%). Infants were found supine or on their sides in 29 (28.4%) and 8 (7.8%) of the cases respectively.

The infants were co-sleeping in 48 (47.1%) deaths and not cosleeping in 54 (52.9%) (Table 3). Co-sleeping was more common in the infants younger than 4-months-old (54.7%) and less common in the older infants (25.9%). In the vast majority of co-sleeping

TABLE 1—Sleeping surface where the infant was found.

Sleeping Surface	All Infants N (%)	$\begin{array}{l} \text{Infants} \leq 4 \text{ Months} \\ N\left(\%\right) \end{array}$	Infants \geq 4 Months N (%)
Adult Mattress	37 (36.3)	28 (37.3)	9 (33.3)
Crib/Bassinet	35 (34.3)	24 (32.0)	11 (40.7)
Couch/Sofa	14 (13.7)	12 (16.0)	2 (7.4)
Water Bed	2 (2.0)	2 (2.7)	0 (0)
Other	14 (13.7)	9 (12.0)	5 (18.5)
Not in Crib/			
Bassinet	67 (65.7)	51 (68.0)	16 (59.3)

TABLE 2—Position in which the infant was found.

Position Found	All Infants N (%)	$\begin{array}{l} \text{Infants} \leq 4 \text{ Months} \\ \text{N}\left(\%\right) \end{array}$	$\begin{array}{l} \text{Infants} \geq 4 \text{ Months} \\ \text{N} (\%) \end{array}$
Prone	63 (61.8)	46 (61.3)	17 (63.0)
Supine	29 (28.4)	20 (26.7)	9 (33.3)
Side	8 (7.8)	8 (10.7)	0 (0)
Other	2 (2.0)	1 (1.3)	1 (3.7)

TABLE 3—Occurrence, number, and identity of co-sleepers.

Co-Sleeping	All Infants N (%)	Infants \leq 4 Months N (%)	Infants ≥ 4 Months N (%)
Yes	48 (47.1)	41 (54.7)	7 (25.9)
No	54 (52.9)	34 (45.3)	20 (74.1)
# Co-Sleepers = 1	24 (50.0)	18 (43.9)	6 (85.7)
# Co-Sleepers = 2	21 (43.8)	20 (48.8)	1 (14.3)
# Co-Sleepers = 3	3 (6.3)	3 (7.3)	0 (0)
Co-Sleeper = Mom	38 (79.2)	34 (82.9)	4 (57.1)
Co-Sleeper = Dad	21 (43.8)	21 (51.2)	0 (0)
Co-Sleeper = Siblings	11 (22.9)	8 (19.5)	3 (42.9)
Co-Sleeper = Other	2 (4.2)	1 (2.4)	1 (14.3)

deaths, the number of co-sleepers with the infant was 1 or 2. The most common individual sleeping with the infant was the mother followed by the father and then siblings.

The preceding data told how frequently any given risk factor was present, but did not reveal how frequently different combinations of risk factors were present. Neither do the data reveal how often infants died without any risk factors. The distribution of risk factors is shown in Fig. 1. When considering all infants, prone sleeping without any other risk factor present was the most common combination, seen in 26 (25.5%). As may be expected, co-sleeping was rare for infants sleeping in a crib or bassinet. The 1 infant co-sleeping in a crib was sharing that sleep surface with 2 siblings. The combination of co-sleeping outside of a crib or bassinet, and not prone was seen in 24 (23.5%). Twenty-three (22.5%) infants exhibited all three risk factors. Not sleeping in a crib or bassinet was rarely present as the only risk factor, seen in 7 (6.9%). In comparison to the infants younger than 4-months-old, the older infants had a notable decrease in the prevalence of co-sleeping either in combination with not sleeping in a crib or bassinet, or in combination with prone sleeping position and not sleeping in a crib or bassinet. The non-co-sleeping older infants, however, were more likely to have slept prone, outside of a crib or bassinet, or a combination of these two.

Only 8 (7.8%) infants exhibited no risk factors. Six of these infants died while sleeping alone, on their back, in a crib or bassinet. Two of these infants died sleeping alone, on their side, in a crib or bassinet. A similar percentage of deaths without risk factors was seen when the data was limited to either infants less than 4-months-old or greater than 4-months-old.

The anatomic diagnoses found at post-mortem examination are summarized in Table 4. The most common diagnoses were pulmonary congestion, hemorrhage, edema, and mediastinal petechiae. The infants less than 4-months-old had a substantially higher frequency of heart malformations than the older infants. The infants greater than 4-months-old were substantially more likely to exhibit pulmonary and tracheal inflammation and neuropathology.

Discussion

We reported the occurrence of unsafe sleep practices in potential cases of SIDS in North Carolina during a 2-year interval. Similar to studies in St. Louis (16) and Alaska (17), we have found that SIDS is uncommon in the absence of a sleep related risk factor. Only 8 (7.8%) deaths were noted in infants sleeping alone, in a crib or bassinet, and on their back or side.

A prone sleeping position was noted in 61.8% of the infants. It has been reported that SIDS deaths in a crib are almost exclusively associated with the prone sleeping position; deaths outside a crib have a lower incidence of prone sleeping position (60%) (17). The current study found that although a majority (77.1%) of infants found in a crib or bassinet were prone, a substantial minority (22.9%) were found supine or on their side. Prone sleeping position was less common in infants found outside of a crib or bassinet (53.7%).

Prone sleep position may pose a risk for SIDS due impaired airway protective reflexes (44), decreased arousability (45), obstructive and mixed sleep apneas (46–48), increased QT interval, and decreased heart rate variability (49). Prone sleep position may also potentiate rebreathing of carbon dioxide when the head is covered with bedding material due to an impaired ability to remove the bedding (50,51). We cannot comment on the role that bedding or head coverage by bedding plays is SIDS since this data was not systematically collected in this study. It is possible that some of the



FIG. 1—Distribution of sleep related risk factors in infants for whom sleep position, sleep surface, and co-sleeping were known. Data are displayed as number (%). Areas of overlap in circles show infants who exhibited the multiple risk factors indicated.

TABLE 4—Anatomic diagnoses.

Diagnosis	All Infants N (%)	Infants ≤ 4 Months N (%)	Infants ≥ 4 Months N (%)
Pulmonary congestion, hemorrhage, or edema	45 (44.1)	34 (45.3)	11 (40.7)
Mediastinal petechiae	45 (44.1)	31 (41.3)	14 (51.9)
Visceral congestion	13 (12.8)	10 (13.3)	3 (11.1)
Heart malformation*	12 (11.8)	11 (14.7)	1 (3.7)
Pulmonary inflammation [†]	11 (10.8)	6 (8.0)	5 (18.5)
Acute or chronic tracheitis	5 (4.9)	2 (2.7)	3 (11.1)
Neuropathology [‡]	5 (4.9)	2(2.7)	3 (11.1)
Aspiration	2(2.0)	0 (0)	2 (7.4)
Fatty liver	2(2.0)	1 (1.3)	1 (3.7)
Hepatitis	1 (1.0)	1 (1.3)	0 (0)
Fluid third spacing	1 (1.0)	1 (1.3)	0 (0)

8 infants in this study without a sleep related risk factor did have their head or face covered. Despite this, we would still conclude that SIDS is rare outside one of the three sleep related risk factors focused on in this study.

Co-sleeping and sleeping outside of a crib or bassinet were noted in 47.1% and 65.7% of the infant deaths, respectively. The majority of non-prone infants were found co-sleeping outside of a crib or bassinet (61.5%) or sleeping alone outside of a crib or bassinet (17.9%). Co-sleeping and/or sleeping outside a crib has been implicated in infant death due to overlaying, entrapment, and suffocation (16,25–27,36,38–40), and some workers have recommended against co-sleeping in adult beds due to these risks (26,39). However, no compelling association between SIDS and co-sleeping has been proven outside the context of a smoking, drinking, or drug abusing parent (11,17,23,24,28–30,37,42,43). Co-sleeping, either in isolation or in combination with another risk factor, was more common in infants less than 4-months-old. Whatever underlying phenomena causes the peak in occurrence of SIDS at 2–4 months old may be exacerbated by co-sleeping in this susceptible age group. It is possible that the lack of certain gross motor skills in younger infants may increase the risk of SIDS while co-sleeping; for example, the majority of infants less than 3-months-old cannot roll over or prop up their chest using their arms (52). Since the current study did not control for tobacco or drug use among co-sleeping adults, we cannot make a generalized recommendation against co-sleeping. However, we can emphasize the observation that SIDS was rare outside one of the three sleep related risk factors examined in this study.

The most common anatomic diagnoses made at post-mortem examination were mediastinal (i.e., heart, lung, or thymus) petechiae and pulmonary congestion, hemorrhage, or edema. The National Institute of Child Health and Human Development Cooperative Epidemiological Study of Sudden Infant Death Syndrome Risk Factors reported that morphologic findings significantly associated with SIDS were thymic petechiae, pleural petechiae, lung congestion and edema, epicardial and myocardial petechiae, and hepatic extramedullary hematopoiesis (53). The diagnoses listed in Table 5 were generally not considered sufficiently severe to account for the infant's death. Some of the diagnoses, cerebral edema for example, were often thought secondary to hypoxic injury and asphyxia.

Although tracheal and pulmonary inflammation were not very common in the SIDS deaths (4.9% and 10.8% of cases, respectively), the significance of these lesions is debatable. A case-control study concluded that upper respiratory and pulmonary inflammation are not a cause of death in SIDS (54). Others, however, have considered acute bronchopneumonia or bronchiolitis to fall into a "gray zone"; these lesions may be considered lethal if sufficiently extensive and severe (53). These distinctions seem to fall into the category of semantics; most pathologists would probably be hesitant in diagnosing SIDS in a case of diffuse severe acute bronchopneumonia. In the cases included in this study, the upper respiratory and pulmonary inflammation were generally not felt to be severe enough to account for death.

The significance of the increased incidence of neuropathology in infants older that 4-months-old remains unclear. Infants who died

of SIDS have been shown to have gliosis in the brainstem and other central nervous system abnormalities (55–57). Altered cardiorespiratory function, arousability, and apoptotic neurodegeneration may follow from these alterations and result in SIDS (57).

A fatty liver was found in 2 (2.0%) infants. Fatty infiltration of the liver is associated with disorders of fatty acid oxidation (FAO) including medium-chain acyl-CoA dehydrogenase deficiency (6,7). These diseases may account for approximately 5% of sudden infant deaths due to fasting intolerance and metabolic decompensation (5,6). A fatty liver is not a sensitive screen for FAO disorders since a third or more of cases may have mild or absent steatosis (6,7). Screening for inborn errors of metabolism may be undertaken by tandem mass spectrometry (58). It is possible that a FAO disorder accounted for some of the deaths in our study population, but, unfortunately, additional confirmatory testing was not systemically undertaken.

Since FAO disorders and prolongation of the QT interval can present as SIDS, we hypothesize that infants with these disorders will be more likely to present without any sleep related risk factors. Given that fatty infiltration of the liver is a marker of FAO disorders, we examined which of the 8 infants identified as sleeping alone, in a crib, and on the back or side were diagnosed with steatosis. Only 1 (12.5%) of these infants had a fatty liver. It is possible that more of these infants had a FAO disorder, since, as discussed previously, steatosis will not be present in all cases. The current data set is too limited to make any conclusive statements with regards to FAO disorders and their relationship to sleep related risk factors. This study also lacks information regarding prolongation of the QT interval. However, future studies might be more able to address these issues. Whether sleep related risk factors potentiate the risk of SIDS in infants with FAO disorders or QT prolongation also remains to be determined.

A limitation of this study was that it was retrospective and uncontrolled. Although we have determined how often sleep related risk factors were present in the study population, we do not know the frequency with which these risk factors are present in the infant population at large. Also, there was the possibility of selection bias so that the sleep surface, sleep position, and occurrence of co-sleeping were more likely to be reported when a risk factor was present. An additional limitation is that we have recorded the position that the infant was in when found. It is possible that the position at the moment of death was different. We also did not systematically determine whether other validated risk factors were present including smoking or substance use by the co-sleeping individual, the use of bulky soft bedding material, and whether bedding material was wrapped around the infant's head. These limitations preclude us from making any definite recommendations with regards to infant sleeping behavior. Certainly our data do not suggest that the prone sleeping position is safe for an infant. Whether or not there is an increased SIDS risk from co-sleeping with an unimpaired parent on an adult mattress requires additional study. Our data do show, however, that SIDS in North Carolina during the study interval was rare for an infant sleeping alone, in a crib, and on its side or back.

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