# **BMJ Open** Comparing asphyxia and unexplained causes of death: a retrospective cohort analysis of sleep-related infant death cases from a state child fatality review programme

Megan Macdonald <sup>(1)</sup>, <sup>1</sup> Daniel Thompson, <sup>2</sup> Robin Perry, <sup>3</sup> Robert Brooks<sup>1</sup>

## ABSTRACT

**Objectives** To examine the characteristics and circumstances of infants who died while sleeping or in a sleep environment and compare deaths classified as either unintentional asphyxia or an unexplained cause. **Design** A retrospective cohort study.

**Setting** Data were extracted from the National Fatality Review Case Reporting System and Florida Vital Statistics databases.

Participants Data on 778 sleep-related infant deaths occurring from 2014 to 2018 in Florida were analysed. Primary outcome measure Cause of death classification as unintentional asphyxia or unexplained.

**Results** Overall, 36% (n=276) of sleep-related infant deaths in this study sample were classified as resulting from an unexplained cause compared with unintentional asphyxia. Most infants were reported to be in an adult bed (60%; n=464) and sharing a sleep surface with a person or animal (60%; n=468); less than half (44%; n=343) were reportedly placed to sleep on their back. After controlling for the influence of other independent variables, female sex (adjusted risk ratio: 1.36; 95% Cl 1.06 to 1.74) and fully obstructed airway condition (adjusted risk ratio: 0.30; 95% Cl 0.18 to 0.50) were associated with an unexplained cause of death.

**Conclusions** The results of this analysis indicate that sleep environment hazards remain prevalent among infants who die suddenly and unexpectedly, regardless of the cause of death determination. While significant differences were observed for some factors, in many others the distributions of both demographic and incident characteristics were similar between unexplained deaths and those resulting from asphyxia. The results of this study support growing evidence that unsafe sleep environments contribute to all forms of sudden unexpected infant death, underscoring the need for standardising cause of death determination practices and promoting consistent, high-quality forensic investigations to accurately explain, monitor and prevent these deaths.

## INTRODUCTION

Sudden unexpected infant death (SUID) remains a leading cause of infant mortality in the USA, accounting for approximately 3600

#### STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ Selection bias was introduced into the study, as the state child welfare agency determines which child deaths will be investigated and therefore case reviewed.
- ⇒ Cause of death information in case reviews is more detailed and informative than relying on International Classification of Diseases 10th Revision (ICD-10) codes alone; however, classifications determined from case reviews are also not directly comparable to standard sudden unexpected infant death categories which are based on ICD-10 codes.
- ⇒ Sample includes only deaths, so true measures of risk of death could not be estimated.

infant deaths every year.<sup>1</sup> SUID refers to the sudden and unexpected death of an infant in which the cause was unclear prior to investigation.<sup>2</sup> SUIDs are classified as either sudden infant death syndrome (SIDS), accidental suffocation and strangulation, or ill-defined and unknown cause based on the International Classification of Diseases 10th Revision (ICD-10) code assigned as the cause of death. Most SUIDs are sleep related or occur during a period of unobserved sleep. In many SUIDs, modifiable risk factors are present in the infant sleeping environment, such as bed sharing, soft bedding and maternal cigarette smoking.<sup>3 4</sup> Despite significant advances in the understanding of SUID epidemiology, unexplained cause of death classifications such as SIDS and other ill-defined and unknown causes of mortality represent most sudden and unexpected infant deaths in recent years.<sup>256</sup>

Rates of SUID declined substantially in the early 1990s following concerted public health campaigns intended to reduce unsafe sleep practices, most notably the American Academy of Pediatrics safe sleep

**To cite:** Macdonald M, Thompson D, Perry R, *et al.* Comparing asphyxia and unexplained causes of death: a retrospective cohort analysis of sleep-related infant death cases from a state child fatality review programme. *BMJ Open* 2022;**12**:e059745. doi:10.1136/ bmjopen-2021-059745

► Prepublication history for this paper is available online. To view these files, please visit the journal online (http://dx.doi. org/10.1136/bmjopen-2021-059745).

Received 02 December 2021 Accepted 28 August 2022

Check for updates

© Author(s) (or their employer(s)) 2022. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

<sup>1</sup>Division of Children's Medical Services, Florida Department of Health, Tallahassee, Florida, USA <sup>2</sup>Independent Statistical Consultant, Tallahassee, Florida, USA

<sup>3</sup>Social Work, Florida Agricultural and Mechanical University, Tallahassee, Florida, USA

#### **Correspondence to**

Megan Macdonald; megan.macdonald@flhealth.gov



recommendations and the commencement of the Back to Sleep campaign. The earlier success of these interventions has diminished and overall SUID rates in the USA have remained relatively unchanged since the late 1990s, fluctuating from 0.95 deaths per 1000 live births from 1998 to 2002, to 0.93 deaths per 1000 live births in 2017.<sup>27</sup> In recent years, SIDS cases have declined while concurrently other SUIDs, due to asphyxia and unknown causes, have increased.<sup>5-9</sup> Currently, unexplained infant deaths classified as SIDS and unknown cause account for, respectively, 38% and 36% of all SUIDs in the USA.<sup>2</sup> The diagnostic shift is thought to be influenced by several factors: stricter adherence to SIDS definitions, a greater understanding of environmental risk factors for SUIDs, improvement in the quality of death scene investigation (DSI), more thorough autopsy practices and influence from child death review teams.<sup>5</sup>

Multiple theories have been proposed to explain possible disease processes or causal pathways underlying SIDS cases, with a particular focus on research into infant vulnerability, but in practice a diagnosis of SIDS is exclusionary in that it is assigned to infant deaths that remain unexplained after a thorough case investigation, including a DSI, autopsy and complete medical history review.<sup>4 12</sup> Moreover, the recent introduction of the term 'SUID' as a specific cause of death used by medical examiners to refer to unexplained sudden infant deaths has further complicated cause of death ascertainment in this area.<sup>12 13</sup> Inconsistent classification of SIDS-like deaths presents a significant challenge to accurate assessment of temporal trends and comparison of SIDS burden geographically, as there is currently no standardised classification system for assigning cause of death in SUIDs and individual medical examiners may certify deaths with similar features quite differently.<sup>5</sup>

Surveillance, monitoring and research into SUID have mostly relied on analysis of Vital Statistics records as these data are widely available and used extensively in the analysis and reporting of public health outcomes. Child fatality review data allow for enhanced study of risk factors and aetiology of sleep-related infant deaths to help explain the circumstances of these incidents and guide prevention efforts.<sup>1415</sup> These data provide for more comprehensive assessment of sleep-related deaths, where Vital Statistics data alone are insufficient to determine environmental risk factors and details from forensic investigations that are captured through the fatality review process. The association between unsafe sleeping environments and SUID risk is well documented, though few studies have examined the relationships between risk factors and specific causes of death in SUID.<sup>3 4 15-18</sup> An analysis of sleep-related infant deaths which used 2008 data from national child fatality review records found that the prevalence of sleep environment risk factors differed significantly across SUID categories.<sup>15</sup> In this study, child death review programme records and death certificate data were used to describe the demographic characteristics, sleep environment risk factors (sleep location, sleep

surface sharing, position placed to sleep, etc) and other incident circumstances for sleep-related infant deaths that occurred in Florida from 2014 to 2018 and to assess the relationship between these factors and the likelihood of an unexplained cause of death.

# METHODS

# Data source

Data for this study were obtained from the National Fatality Review Case Reporting System (NFR-CRS) and Florida Vital Statistics infant death records linked to birth records. The NFR-CRS is a national web-based system for collection and analysis of child death review data where local review teams enter case information after completing death reviews. The Florida Child Abuse Death Review (CADR) programme uses local committees to conduct detailed reviews of the facts and circumstances of child deaths reported to the Florida Abuse Hotline on suspicion of abuse or neglect, regardless of the final determination of case maltreatment status. Data from completed case reviews are then entered into the NFR-CRS. The system's data elements include demographic information on the child, the location and setting of the incident, manner, and cause of death, known risk factors related to the sleep environment and other risk factors associated with infant death. Independent variables were selected according to their relevance to sleep-related infant death and cause of death classification based on review of the literature. In the planning phase of this analysis, a high proportion of unknown race/ethnicity records was observed in the study sample from the NFR-CRS (9.5%) compared with the same fields for matching records in Vital Statistics (2.6%). Therefore, demographic information on the infant was obtained from Vital Statistics while characteristics of the death incident, including sleep environment factors as well as other death investigation information, were acquired from case review records.

#### **Study sample**

Cases of sleep-related infant death were identified in the NFR-CRS among infants less than 12 months old that occurred in Florida between 2014 and 2018, where the death was determined by case review to be related to sleeping or the sleep environment. NFR-CRS records were then matched to corresponding linked death-tobirth records; the birth certificate data fields were null 3.7% of matched vital records, which is likely explained by deaths that occurred in non-resident children.

A total of 825 sleep-related infant deaths met inclusion criteria for the study. Records with an explained medical condition or injury indicated as the cause of death in the NFR-CRS record (n=34) or with a reported manner of death that was inconsistent with SUIDs, such as homicide (n=4), were excluded. Sleep-related infant deaths that occurred in 2019 were excluded from the study due to limited cases with complete data entry available for analysis (n=9), resulting in a final study sample of 778 deaths.



**Figure 1** Participant flow chart. NFR-CRS, National Fatality Review Case Reporting System.

The flow chart in figure 1 illustrates the process followed to select participants. Each record was assigned to one of two causes of death categories: unintentional asphyxia and unexplained cause; the unexplained group included deaths where the primary cause of death, as recorded in the NFR-CRS, was undetermined, unknown, SIDS or 'SUID'. In this analysis, 'SUID' refers to a cause of death used by medical examiners to certify some unexplained infant deaths and is distinct from the overarching SUID reporting categories based on ICD-10 codes.

Demographic characteristics analysed in the study were infant age category, race and ethnicity, sex, maternal age and maternal education. Selected birth outcomes, including gestational age of the infant in weeks and birth weight in grams, were also included.

Independent variables in the analysis reflecting the circumstances of sleep-related deaths were created using items from the NFR-CRS detailing sleep environment risk factors and the place and position in which the infants were found. Sleep environment risk factors included the sleeping location or type of place where the infant was found unresponsive, whether the infant was sleeping on the same sleep surface with a person or animal, the position in which the infant was put to sleep and the position of the infant when found unresponsive.

Level of airway obstruction was determined by the condition of the infant's nose, mouth and/or chest when

they were found. If, when the infant was found, their nose and mouth were completely covered by an object, or their chest or neck was fully compressed, thereby preventing the lungs from expanding, the airway was described as fully obstructed. Likewise, if only part of the nose or mouth was covered, or only part of the chest or neck was compressed, or where the infant's position compressed the airway, the infant's airway was described as partially obstructed. The airway was categorised as unobstructed if nothing was covering the nose or mouth or compressing the neck or chest.

Other variables related to the incident used in the analysis were the place where the sleep-related death incident occurred, the type of area (urban, suburban/rural, unknown) where the incident occurred, whether a DSI was conducted at the place of the incident and the year the death occurred.

The Florida Department of Health Institutional Review Board (IRB) office determined that this study was exempt from review.

## Patient and public involvement

The public was not involved in the design, or conduct, or reporting or dissemination plans of this research, as it was not appropriate or possible to do so. As the study was a retrospective analysis performed using an existing data set, the relevant ethical governance committee determined that this study was exempt from IRB review.

#### **Data analysis**

All data analyses were performed using Stata V.15. Descriptive statistics were computed for all demographic and incident characteristics and were stratified by the two causes of death categories: unintentional asphyxia and unexplained cause. Pearson's  $\chi^2$  tests were performed to determine whether cause of death category was significantly associated with any of the independent variables. Univariate and multivariate negative binomial regression models were used to calculate unadjusted and adjusted risk ratios (aRR) to assess the relationship between all independent variables and cause of death category. Variance inflation factor and correlation analyses were performed on the adjusted regression model to assess multicollinearity among the selected independent variables. Results indicated that low to moderate correlation was present only in a few variables and was unlikely to affect the multivariate regression results. Five of the independent variables of interest contained missing values, for which the range of missing data across these variables did not exceed 3.7% of total observations in the data set. Missing data, where they exist, are displayed in the variable frequency distributions but have been excluded from  $\chi^2$  and regression analyses.

## RESULTS

From 2014 to 2018, there were 1068 SUID deaths recorded in Florida and the sample of sleep-related infant

deaths in this study included approximately 778 (73%) of total SUIDs in Florida during this period. Of the 778 sleep-related infant deaths included in this analysis, the majority were male (59%) and less than 5 months old (77%). Non-Hispanic white infants accounted for 35% of these deaths, whereas 38% were non-Hispanic black, 17% were Hispanic and the remaining 10% comprised other and unknown races. Nearly 20% of these infants had low birth weight, defined as weight less than 2500 g at birth, and 20% were born preterm, defined as less than 37 weeks' gestation. More than half (52%) of the infants were born to mothers aged 25 and younger and the majority (65%) were born to mothers with a high school or lower education.

Most deaths (76%) occurred in the infant's home. Forty per cent took place in an urban area, whereas 41%were in a rural or suburban area, and in 19% of cases the type of area where the incident occurred was unknown. Sixty per cent of infants were sleeping in an adult bed at the time of death. Only 17% were found sleeping in a crib or bassinet and 11% were sleeping on a couch or chair. Regardless of sleep location, 60% of infants were sharing a sleep surface with a person or animal. Forty-four per cent of infants were reported to be placed to sleep on their backs, while 39% were reportedly placed on their side or stomach, and in 17% of cases, sleep placement was unknown. At the time of death, most infants (60%)were found on their side or stomach. In 30% of deaths, the infant's airway was fully obstructed by a person or object, whereas 19% of infants were found to have partial or no airway obstruction. Airway condition was unknown or missing in slightly over half (51%) of the deaths. Frequency distributions and proportions of deaths by cause of death category and infant and incident characteristics are displayed in tables 1 and 2.

Based on  $\chi^2$  analysis of all independent variables, infant sex, type of area, sleep location, sleep surface sharing, sleep position when found, airway condition, performance of a DSI and the year of death were significantly associated with cause of death category.

Table 3 presents the results of unadjusted and adjusted negative binomial regression models comparing deaths classified as unexplained with deaths classified as unintentional asphyxia.

Univariate analysis found that infant sex was the only infant demographic characteristic that was significantly associated with unexplained cause of death. Among incident characteristics, univariate models showed significant risk ratios for type of area and DSI, but these factors lost statistical significance after adjusting for other variables. In the final adjusted regression model, infant sex, airway condition and year of death remained significantly associated with unexplained cause of death. Female infants were over 30% more likely to have an unexplained cause of death (aRR 1.36; 95% CI 1.06 to 1.74) and deaths in which the infant had a fully obstructed airway were 30% as likely to be classified as unexplained (aRR 0.30; 95% CI 0.18 to 0.50). Deaths that occurred in 2017 were 64% as likely as those that occurred in 2014 to be unexplained (aRR 0.64; 95% CI 0.42 to 0.96).

# DISCUSSION

This analysis of NFR-CRS data showed that the prevalence of known sleep environment risk factors among sleeprelated infant deaths remains high; only 17% of infants were sleeping in a crib or bassinet when found and less than half (44%) were reportedly placed to sleep on their backs. Notably, most infants in the study group were found in an adult bed (60%) and sharing a sleep surface with another person or animal (60%). Findings from this study are consistent with other research showing that cause of death classification varies based on the presence of risk factors in the sleep environment.<sup>15</sup> The general high prevalence of sleep environment risk factors across different cause of death categories in this study also aligns with other studies which have used child death review data to examine SUIDs.<sup>1415</sup>

One of the most striking results from the analysis of demographic characteristics in this study population is the association between infant sex and cause of death classification; while most of sleep-related deaths were among male infants, deaths among females were significantly more likely to be unexplained compared with their male counterparts. There is a well-documented sex disparity in SIDS cases, with male infants accounting for 60% of SIDS deaths, but evidence from current medical literature does not explain the relationship between sex and unexplained cause of death compared with asphyxia death that was observed in this study.<sup>19</sup> Previous studies on SUID classification have found significant variation in cause of death between different racial/ethnic and age groups, but not by sex<sup>15</sup>; conversely, demographic features across asphyxia and unexplained death categories in this study revealed remarkably similar race/ethnicity and age distributions between the outcome groups.

The variable most significantly associated with cause of death was condition of the infant's airway; deaths in which the infant had a fully obstructed airway were less likely to be unexplained compared with infants who had an unobstructed airway. This is not a surprising finding, as medical examiners rely heavily on physical evidence of asphyxia to certify the death as suffocation.<sup>12 20</sup> Interestingly, however, there was no difference in the cause of death category for infants with partial or unknown airway obstruction; moreover, infants with full airway obstruction still accounted for 12% of unexplained deaths.

Infant deaths that are sudden and unwitnessed often fall into a medical 'gray area' for forensic pathologists where the lack of a standard approach for sleep-related infant death classification and variation in the way individual medical examiners consider evidence further compound inconsistencies in SUID classification.<sup>5 6 12 21</sup> Importantly, studies have shown that when pathologists applied standardised criteria for SIDS certification in a reclassification exercise of SUIDs, natural causes of death were significantly reduced.<sup>10</sup> Establishing clear definitions and classification

Characteristic	Total (n=778)	Unexplained* (n=276)	Unintentional asphyxia (n=502)
Less than 2 months	232 (29.8)	86 (31 2)	146 (29 1)
2–4 months	369 (47 4)	130 (47 1)	239 (47.6)
5–11 months	177 (22.8)	60 (21 7)	117 (23.3)
Bace/ethnicity (n_%)	(111 (1210)	00 (2)	
White, non-Hispanic	269 (34.6)	93 (33.7)	176 (35.1)
Black, non-Hispanic	299 (38.4)	104 (37.7)	195 (38.8)
Hispanic	135 (17.4)	49 (17.8)	86 (17.1)
Other, non-Hispanic	55 (7.1)	23 (8.3)	32 (6.4)
Unknown	20 (2.6)	7 (2.5)	13 (2.6)
Sex (n. %)		. ()	
Male	458 (58.9)	145 (52.5)	313 (62.4)
Female	320 (41.1)	131 (47.5)	189 (37.7)
Gestational age in weeks (n. %)			
≥37	593 (76.2)	206 (74.6)	387 (77.1)
<37	155 (19.9)	58 (21.0)	97 (19.3)
Unknown	1 (0.1)	1 (0.4)	0 (0)
Missing	29 (3.7)	11 (4.0)	18 (3.6)
Birth weight in grams (n, %)			
≥2500	629 (80.9)	219 (79.4)	410 (81.7)
<2500	149 (19.2)	57 (20.7)	92 (18.3)
Maternal age (n, %)			
<20	86 (11.1)	25 (9.1)	61 (12.2)
20–25	320 (41.1)	113 (40.9)	207 (41.2)
26–35	293 (37.7)	104 (37.7)	189 (37.7)
>35	50 (6.4)	23 (8.3)	27 (5.4)
Missing	29 (3.7)	11 (4.0)	18 (3.6)
Maternal education level (n, %)			
Less than high school	189 (24.3)	68 (24.6)	121 (24.1)
High school graduate	318 (40.9)	97 (35.1)	221 (44.0)
Some college	186 (23.9)	80 (29.0)	106 (21.1)
College graduate	45 (5.8)	16 (5.8)	29 (5.8)
Unknown	11 (1.4)	4 (1.5)	7 (1.4)
Missing	29 (3.7)	11 (4.0)	18 (3.6)

\*Includes cases with undetermined, unknown, sudden infant death syndrome (SIDS) and sudden unexpected infant death (SUID) as the underlying cause documented on the death certificate.

criteria for these types of infant deaths could reduce inconsistencies in cause of death certification and improve the accuracy of SUID surveillance and mitigation efforts.

A DSI was performed in 87% of the cases, where the proportion was slightly lower in unexplained deaths compared with asphyxia deaths. Performance of a DSI was not significantly associated with cause of death, but this is possibly due to small numbers in the reference group (n=34), as there were relatively few cases that did not have

a DSI. Also, there may be wide variation in the quality and components of DSIs that were not explored in this analysis.

There are several important limitations to this study. The state's child welfare agency determines which child deaths are investigated on suspicion of maltreatment and referred to the CADR programme for review, and therefore cases in the NFR-CRS may not be representative of all sleeprelated infant deaths or SUIDs as this element introduced selection bias into the study. Additionally, some cases may

Table 2 Incident characteristics of sleep-related infant deaths by cause of death category					
Characteristic	Total (n=778)	Unexplained* (n=276)	Unintentional asphyxia (n=502)		
Place of death (n, %)					
Child's home	592 (76.1)	214 (77.5)	378 (75.3)		
Friend or relative's home	144 (18.5)	43 (15.6)	101 (20.1)		
Other	42 (5.4)	19 (6.9)	23 (4.6)		
Type of area (n, %)					
Urban	308 (39.6)	125 (45.3)	183 (36.5)		
Suburban or rural	322 (41.4)	98 (35.5)	224 (44.6)		
Unknown	148 (19.0)	53 (19.2)	95 (18.9)		
Sleep location (n, %)					
Crib/bassinet	132 (17.0)	56 (20.3)	76 (15.1)		
Adult bed	464 (59.6)	157 (56.9)	307 (61.2)		
Couch/chair	84 (10.8)	23 (8.3)	61 (12.2)		
Other	93 (12.0)	36 (13.0)	57 (11.4)		
Unknown	5 (0.6)	4 (1.5)	1 (0.2)		
Sharing sleep surface (n, %)					
No	299 (38.4)	120 (43.5)	179 (35.7)		
Yes	468 (60.2)	150 (54.4)	318 (63.4)		
Unknown	11 (1.4)	6 (2.2)	5 (1.0)		
Position placed to sleep (n, %)					
On back	343 (44.1)	128 (46.4)	215 (42.8)		
On side/stomach	300 (38.6)	90 (32.6)	210 (41.8)		
Unknown	135 (17.4)	58 (21.0)	77 (15.3)		
Position when found (n, %)					
On back	160 (20.6)	66 (23.9)	94 (18.7)		
On side/stomach	464 (59.6)	151 (54.7)	313 (62.4)		
Unknown	154 (19.8)	59 (21.4)	95 (18.9)		
Airway condition (n, %)					
Unobstructed	77 (9.9)	34 (12.3)	43 (8.6)		
Fully obstructed	237 (30.5)	34 (12.3)	203 (40.4)		
Partially obstructed	70 (9.0)	33 (12.0)	37 (7.4)		
Unknown	372 (47.8)	170 (61.6)	202 (40.2)		
Missing	22 (2.8)	5 (1.8)	17 (3.4)		
Death scene investigation (n, %)					
Yes	677 (87.0)	227 (82.3)	450 (89.6)		
No	34 (4.4)	14 (5.1)	20 (4.0)		
Unknown	43 (5.5)	23 (8.3)	20 (4.0)		
Missing	24 (3.1)	12 (4.4)	12 (2.4)		
Year of death (n, %)					
2014	151 (19.4)	61 (22.1)	90 (17.9)		
2015	153 (19.7)	59 (21.4)	94 (18.7)		
2016	155 (19.9)	58 (21.0)	97 (19.3)		
2017	175 (22.5)	45 (16.3)	130 (25.9)		
2018	144 (18.5)	53 (19.2)	91 (18.1)		

\*Includes cases with undetermined, unknown, sudden infant death syndrome (SIDS) and sudden unexpected infant death (SUID) as the underlying cause documented on the death certificate.

Table 3 Unadjusted and adjusted risk ratios of unexplained death (compared with unintentional asphyxia deaths)				
Characteristic	RR (95% CI)	aRR (95% CI)*		
Age (months)				
<2	Reference	Reference		
2–4	0.95 (0.72 to 1.25)	1.05 (0.77 to 1.43)		
5–11	0.91 (0.66 to 1.27)	1.05 (0.71 to 1.56)		
Race/ethnicity				
White, non-Hispanic	Reference	Reference		
Black, non-Hispanic	1.01 (0.76 to 1.33)	1.05 (0.76 to 1.46)		
Hispanic	1.05 (0.74 to 1.48)	1.03 (0.70 to 1.50)		
Other, non-Hispanic	1.21 (0.77 to 1.91)	1.19 (0.72 to 1.96)		
Unknown	1.01 (0.47 to 2.18)	1.42 (0.64 to 3.18)		
Gestational age (weeks)				
≥37	Reference	Reference		
<37	1.08 (0.80 to 1.44)	0.98 (0.65 to 1.47)		
Unknown	2.88 (0.40 to 20.53)	5.21 (0.64 to 42.23)		
Birth weight (g)				
≥2500	Reference	Reference		
<2500	1.10 (0.82 to 1.47)	1.13 (0.75 to 1.71)		
Maternal age				
<20	Reference	Reference		
20–25	1.21 (0.79 to 1.87)	1.22 (0.76 to 1.94)		
26–35	1.22 (0.79 to 1.89)	1.12 (0.69 to 1.83)		
>35	1.58 (0.90 to 2.79)	1.44 (0.77 to 2.70)		
Maternal education level				
Less than high school	Reference	Reference		
High school graduate	0.84 (0.62 to 1.16)	0.78 (0.56 to 1.10)		
Some college	1.20 (0.86 to 1.65)	1.14 (0.80 to 1.64)		
College graduate	0.99 (0.57 to 1.70)	0.84 (0.47 to 1.51)		
Unknown	1.04 (0.60 to 1.82)	1.14 (0.40 to 3.26)		
Sex				
Male	Reference	Reference		
Female	1.29 (1.02 to 1.64)	1.36 (1.06 to 1.76)		
Place of death				
Child's home	Reference	Reference		
Friend or relative's home	0.83 (0.60 to 1.15)	0.87 (0.60 to 1.26)		
Other	1.25 (0.78 to 2.00)	0.93 (0.53 to 1.63)		
Type of area				
Urban	Reference	Reference		
Suburban or rural	0.75 (0.58 to 0.98)	0.78 (0.58 to 1.04)		
Unknown	0.88 (0.64 to 1.22)	0.77 (0.51 to 1.16)		
Sleep location				
Crib/bassinet	Reference	Reference		
Adult bed	0.80 (0.59 to 1.08)	1.05 (0.68 to 1.63)		
Couch/chair	0.65 (0.40 to 1.05)	1.06 (0.65 to 1.74)		
Other	0.91 (0.60 to 1.39)	0.90 (0.56 to 1.43)		
Unknown	1.89 (0.68 to 5.20)	2.32 (0.76 to 7.04)		
Sharing sleep surface				
No	Reference	Reference		

Continued

Table 5 Continued				
Characteristic	RR (95% CI)	aRR (95% CI)*		
Yes	0.80 (0.63 to 1.02)	0.77 (0.53 to 1.11)		
Unknown	1.36 (0.60 to 3.09)	0.68 (0.23 to 2.04)		
Position placed to sleep				
On back	Reference	Reference		
On side/stomach	0.80 (0.61 to 1.05)	0.87 (0.62 to 1.21)		
Unknown	1.15 (0.84 to 1.57)	1.02 (0.69 to 1.51)		
Position when found				
On back	Reference	Reference		
On side/stomach	0.79 (0.59 to 1.05)	0.91 (0.63 to 1.31)		
Unknown	0.93 (0.65 to 1.32)	0.78 (0.51 to 1.21)		
Airway condition				
Unobstructed	Reference	Reference		
Fully obstructed	0.32 (0.20 to 0.52)	0.28 (0.16 to 0.49)		
Partially obstructed	1.07 (0.66 to 1.72)	0.92 (0.53 to 1.59)		
Unknown	1.03 (0.71 to 1.50)	0.99 (0.65 to 1.52)		
Death scene investigation performed				
Yes	Reference	Reference		
No	1.23 (0.72 to 2.10)	1.24 (0.70 to 2.20)		
Unknown	1.60 (1.04 to 2.45)	1.45 (0.89 to 2.39)		
Year of death				
2014	Reference	Reference		
2015	0.95 (0.67 to 1.37)	0.97 (0.65 to 1.46)		
2016	0.93 (0.65 to 1.33)	0.96 (0.65 to 1.44)		
2017	0.64 (0.43 to 0.94)	0.65 (0.42 to 0.99)		
2018	0.91 (0.63 to 1.32)	0.91 (0.60 to 1.38)		
*Adjusted for all independent variables in this table				

"Adjusted for all independent variables in this tabl

aRR, adjusted risk ratio; RR, risk ratio.

have pending investigations or legal proceedings which delay timely data entry. Furthermore, as the primary cause of death documented in the NFR-CRS was used in analysis rather than ICD-10 codes, the estimates of sleep-related infant death in this study are not directly comparable to SUID estimates that are generated from Vital Statistics records. While this is represented as a limitation, it may also reflect a strength of the study, in that fatality reviews offer more granular information on cause of death than what is available in the Vital Statistics records. ICD-10 codes have limitations regarding cause of death in SUID, as the assigned ICD-10 code does not always reflect what the medical examiner intended during death certification. For example, a death which the medical examiner indicated was caused by 'SUID' would ultimately be coded as SIDS based on National Center for Health Statistics' ICD-10 criteria. Therefore, this sample of sleep-related infant deaths may not be fully representative of all SUIDs that occurred in the state during the same period.

Exclusion of infant death records which were determined to be sleep related by review teams but had a medical condition indicated as the primary cause of death also presents a limitation in that some of these deaths may have been subject to misclassification in the role the sleep environment played in contributing to the death. Furthermore, local review teams may ascertain cause of death differently, as the National Fatality Review Case Reporting Form allows committees to select primary causes of death that may not match the immediate cause of death indicated on the death certificate. The majority of sleep-related infant deaths in this study were due to asphyxia; by contrast, infant death records in Florida reveal that, during the same time period, 56% of all SUID cases in the state were classified as SIDS or an ill-defined/unknown cause of death.22 Additionally, due to the nature of the NFR-CRS, only information on deceased infants was available for analysis, precluding the ability to determine aetiological relationships between documented sleep environment factors and the risk of SUID or differential causes of death. The authors did not have access to meetings, recordings and/or minutes of these meetings from which contextual factors could be ascertained to determine their potential influence on observed differences (and level of inter-rater/committee reliability) in coding practices. In the absence of reliability checking, there exists potential for bias in the assessment criteria used between individual review teams that may also differ from child fatality review team approaches in other states and abroad.

Decades of research and large-scale public health interventions have led to improved understanding of SUID aetiology and more effective prevention strategies, vet SUID rates have plateaued in recent years and most of these deaths are still unexplained. Thorough and consistent DSIs are critical for the accurate assessment of sleep-related infant death, as most of these deaths are unwitnessed and autopsy results alone are usually insufficient to explain how the deaths were caused.<sup>23</sup> Further, inconsistencies in the quality and appraisal of forensic evidence and thus the classification of sleep-related deaths compound the challenges of accurately describing and communicating this problem to the public and professionals in fields of medicine and public health. Implementation of a standard approach to SUID classification and improvement in the quality and consistency of DSIs are necessary to improve SUID monitoring and reporting and thus the ability to accurately explain these deaths and guide evidence-based prevention efforts.

**Contributors** MM and RB conceptualised and designed the study, carried out the initial analysis, drafted the initial manuscript, and reviewed and revised the manuscript. MM performed the final analysis. DT advised the team on statistical analysis methods and reviewed the manuscript. RP critically reviewed the manuscript for important intellectual content. RB acted as guarantor of the study. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

**Funding** This publication was supported by the Health Resources and Services Administration (HRSA) of the US Department of Health and Human Services (HHS) as part of the State Systems Development Initiative (SSDI) award (6H18MC00010-24-01) totalling \$100 000, in association with the Title V Block Grant.

**Disclaimer** The contents are those of the authors and do not necessarily represent the official views of, nor an endorsement by, HRSA, HHS or the US Government.

Competing interests None declared.

Patient and public involvement Patients and/or the public were not involved in the design, or conduct, or reporting or dissemination plans of this research.

Patient consent for publication Not applicable.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement No data are available.

**Open access** This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: http://creativecommons.org/licenses/by-nc/4.0/.

#### **ORCID iD**

Megan Macdonald http://orcid.org/0000-0002-8842-2974

#### REFERENCES

1 United States Department of Health and Human Services (US DHHS) CoDCaPC, National Center for Health Statistics (NCHS), Division of Vital Statistics (DVS). Linked birth / infant death records 2007-2017, as compiled from data provided by the 57 vital statistics jurisdictions through the vital statistics cooperative program, on CDC wonder on-line database 2019. Available: http://wonder.cdc.gov/lbd-current. html [Accessed 03 Sep 2019].

- 2 Centers for Disease Control and Prevention (CDC) DoRH. National Center for Chronic Disease Prevention and Health Promotion Sudden Unexpected Infant Death and Sudden Infant Death Syndrome 2018. Available: https://www.cdc.gov/sids/about/index. htm?CDC\_AA\_refVal=https%3A%2F%2Fwww.cdc.gov%2Fsids% 2FAboutSUIDandSIDS.htm [Accessed 03 Oct 2019].
- 3 Task Force On Sudden Infant Death S. SIDS and other sleep-related infant deaths: updated 2016 recommendations for a safe infant sleeping environment. *Pediatrics* 2016;138.
- 4 Moon RY, Darnall RA, Feldman-Winter L, TASK FORCE ON SUDDEN INFANT DEATH SYNDROME. Sids and other sleep-related infant deaths: evidence base for 2016 updated recommendations for a safe infant sleeping environment. *Pediatrics* 2016;138.
- 5 Shapiro-Mendoza CK, Parks SE, Brustrom J, et al. Variations in cause-of-death determination for sudden unexpected infant deaths. *Pediatrics* 2017;140. doi:10.1542/peds.2017-0087. [Epub ahead of print: 05 06 2017].
- 6 Shapiro-Mendoza CK, Camperlengo L, Ludvigsen R, et al. Classification system for the sudden unexpected infant death case registry and its application. *Pediatrics* 2014;134:e210–9.
- 7 Shapiro-Mendoza CK, Parks S, Lambert AE. The Epidemiology of Sudden Infant Death Syndrome and Sudden Unexpected Infant Deaths: Diagnostic Shift and other Temporal Changes. In: Duncan JR, Byard RW, eds. SIDS sudden infant and early childhood death: the past, the present and the future. Adelaide: University of Adelaide Press (c) 2018 The Contributors, with the exception of which is by Federal United States employees and is therefore in the public domain, 2018.
- 8 Shapiro-Mendoza CK, Camperlengo LT, Kim SY, et al. The sudden unexpected infant death case registry: a method to improve surveillance. *Pediatrics* 2012;129:e486–93.
- 9 Matshes EW, Lew EO. An approach to the classification of apparent asphyxial infant deaths. *Acad Forensic Pathol* 2017;7:200–11.
- 10 Shipstone RA, Young J, Thompson JMD, et al. An evaluation of pathologists' application of the diagnostic criteria from the San Diego definition of SIDS and unclassified sudden infant death. Int J Legal Med 2020;134:1015–21.
- 11 Byard RW, Krous HF. Research and sudden infant death syndrome: definitions, diagnostic difficulties and discrepancies. J Paediatr Child Health 2004;40:419–21.
- 12 Hunt CE, Darnall RA, McEntire BL, et al. Assigning cause for sudden unexpected infant death. Forensic Sci Med Pathol 2015;11:283–8.
- 13 Goldstein RD, Blair PS, Sens MA, et al. Inconsistent classification of unexplained sudden deaths in infants and children hinders surveillance, prevention and research: recommendations from the 3rd international Congress on sudden infant and child death. Forensic Sci Med Pathol 2019;15:622–8.
- 14 Brixey SN, Kopp BC, Schlotthauer AE, *et al*. Use of child death review to inform sudden unexplained infant deaths occurring in a large urban setting. *Inj Prev* 2011;17 Suppl 1:i23–7.
- 15 Schnitzer PG, Covington TM, Dykstra HK. Sudden unexpected infant deaths: sleep environment and circumstances. *Am J Public Health* 2012;102:1204–12.
- 16 Erck Lambert AB, Parks SE, Cottengim C, *et al.* Sleep-Related infant suffocation deaths attributable to soft bedding, overlay, and wedging. *Pediatrics* 2019;143.
- 17 Maged M, Rizzolo D. Preventing sudden infant death syndrome and other sleep-related infant deaths. JAAPA 2018;31:25–30.
- 18 Sauber-Schatz EK, Sappenfield WM, Shapiro-Mendoza CK. Comprehensive review of sleep-related sudden unexpected infant deaths and their investigations: Florida 2008. *Matern Child Health J* 2015;19:381–90.
- 19 Mage DT. Donner M. a unifying theory for SIDS. Int J Pediatr 2009;2009:368270.
- 20 Garstang J, Ellis C, Griffiths F, et al. Unintentional asphyxia, SIDS, and medically explained deaths: a descriptive study of outcomes of child death review (CDR) investigations following sudden unexpected death in infancy. *Forensic Sci Med Pathol* 2016;12:407–15.
- 21 Corey TS, Hanzlick R, Howard J, et al. A functional approach to sudden unexplained infant deaths. Am J Forensic Med Pathol 2007;28:271–7.
- 22 Health FDo. Florida Community Health Assessment Resource Tool Set (FIHealthCHARTS), 2019. Available: http://www.flhealthcharts. com [Accessed 03 Sep 2019].
- 23 Erck Lambert AB, Parks SE, Camperlengo L, *et al.* Death scene investigation and autopsy practices in sudden unexpected infant deaths. *J Pediatr* 2016;174:84–90.