Sudden Unexpected Infant Death Certification in New York City: Intra-Agency Guideline Compliance and Variables that May Influence Death Certification

Melissa A. Pasquale-Styles, Margaret Regensburg, Ruijun Bao

ABSTRACT

Introduction: Differences in certification of similar sudden infant deaths exists among forensic pathologists. This study sought to measure adherence to intra-agency guidelines for infant death certification in one jurisdiction and describe variables that may be associated with the differentiation of sudden infant death syndrome (SIDS), asphyxia, and undetermined death certifications.

Methods: A retrospective study of deaths at the New York City Office of Chief Medical Examiner identified 427 sudden infant deaths with investigation and autopsy whose cause of death was ruled SIDS, asphyxia, or undetermined. Cases were reviewed for number and types of risk factors for asphyxia, demonstrable evidence of asphyxia, potential competing causes of death, and a doll reenactment. Descriptive and statistical analysis was performed.

Results: Of 427 deaths, the causes of 100 deaths (23.4%) were ruled asphyxia; 43 (10.0%) SIDS; and 284 (66.5%) undetermined. Forensic pathologists conformed to agency guidelines in 406 deaths (95.1%). Three or more risk factors for asphyxia were found in 328 deaths (76.8%). Demonstrable evidence of asphyxia (40.7%) was most associated with a certification of asphyxia. A potential competing cause of death (20%) was most associated with undetermined. A doll reenactment had little association with certification type.

Discussion: Guidelines in one agency were effective at limiting incorrect SIDS diagnoses. The interpretation of risk factors can be subjective. Diagnostic overlap occurred in deaths certified differently as SIDS, asphyxia, and undetermined, despite similar findings. Elimination of SIDS as a certification option and better guidelines that help differentiate asphyxia and undetermined deaths are recommended for improved infant death certification. *Acad Forensic Pathol.* 2017 7(4): 536-550

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INFORMATION

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INTRODUCTION

Following the inception of sudden infant death syndrome (SIDS) in 1969, there was a trend to certify many sudden infant deaths as SIDS through the late 1990s (1, 2). The incidence of sudden infant deaths declined in the late twentieth century, following changing infant sleep practices to favor supine sleep (3). Supine sleep was formally promoted by the American Academy of Pediatrics' safe sleep recommendations in 1992 and the Back to Sleep campaign in 1994 (4-6). While the incidence of sudden infant deaths of all causes has relatively plateaued since 1998, a diagnostic shift has occurred since the early 2000s in which fewer deaths are attributed to SIDS and more are ascribed to unsafe sleep-related asphyxia or undetermined causes (1, 3, 6, 7).

This diagnostic shift has occurred primarily due to better investigations and awareness of the circumstances surrounding infant deaths (3). Improved death scene investigations have found one or more unsafe sleep-related risk factors for asphyxia in most sudden infant deaths (8, 9). These include unapproved infant sleep surfaces such as adult beds or couches, soft sleep surfaces, bed sharing, prone or side position, and soft objects or loose bedding in the sleep space (10-12). Sudden infant death syndrome is often assigned a natural manner of death by convention, and the certification of SIDS has lost favor among many certifiers who prefer variations of asphyxial causes of death or undetermined when extrinsic unsafe sleep factors are determined to have caused the death or cannot be excluded (8, 13).

This diagnostic shift has not been geographically uniform throughout the United States (2). Infant death investigation is performed by medical examiners and coroners (ME/C) with independent jurisdiction, variable training and/or quality of investigation, and wide latitude for judgment in cause and manner of death certification. There is no single method of infant death investigation or interpretation of the findings, and there is disagreement among ME/C about the circumstances under which SIDS is an acceptable cause of death, if the term should be used at all, and what

unsafe sleep environmental factors are sufficient to determine an asphyxial death. The differences in death certification practice are evident when comparing causes of infant death between jurisdictions around the United States (2, 14), but variations can also exist among medical examiners working within the same office (based on the experience of two of the four authors). The current state of inconsistent infant death certification practices results in confusion for epidemiologists, families, clinicians, researchers, and advocacy groups (15, 16).

The definition of SIDS that is recognized to date at the New York City Office of Chief Medical Examiner (NYC OCME) as an acceptable cause of select deaths was proposed by Willinger et al. in 1989 (published in 1991), and is:

The sudden death of an infant under one 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 (17).

With recognition of risk factors for infant asphyxia, former Chief Medical Examiner of the NYC OCME, Dr. Charles S. Hirsch, issued internal agency guidelines circa 2002 for the use of SIDS in infant deaths in which the autopsy and associated postmortem studies fail to disclose an etiology. These guidelines are still included in the agency's training manual for newly hired forensic pathologists.

The guidelines specify that in order to certify a death as SIDS, perinatal and clinical history must be obtained and a death scene investigation must be done for the purpose of identifying environmental hazards that may cause or contribute to the death. The guidelines explicitly disqualify SIDS as the cause of death if a scene investigation is not performed, the infant is bed sharing with adults or siblings, the infant dies while sleeping on soft compressible bedding, or the death occurs during prone sleep on anything other than a firm mattress designed for infant safety. In cases where SIDS-disqualifying criteria exist, the medical examiner has the option to choose asphyxia due to

an underlying unsafe sleep-related cause (herein referenced only as “asphyxia”) or undetermined as the cause of death.

These intra-agency guidelines set forth by Dr. Hirsch only specify when SIDS cannot be used and do not require a SIDS certification if the death qualifies as such based on the guidelines. They do not disqualify prone position alone from SIDS certification despite research supporting prone and side position as risk factors for sudden infant death and association with asphyxia (10, 11, 18). The guidelines also do not disqualify SIDS in merely the presence of soft objects or loose bedding though these have also been identified as unsafe sleep factors by the American Academy of Pediatrics (18). Likewise, a doll reenactment is not required to certify a death as SIDS though doll reenactments are strongly advised in all sudden infant death investigations as a matter of routine practice at the NYC OCME.

Within these guidelines, medical examiners retain individual discretion to exercise their professional judgment in assessing each death independently and determining cause and manner of death. Opportunities to reinforce the guidelines and provide feedback to medical examiners occur during regular internal death certificate reviews, internal case conferences, and multidisciplinary child fatality review conferences.

This study had two parts. First, it sought to measure medical examiners’ compliance with intra-agency guidelines for SIDS certification in one jurisdiction. Second, it separately sought to determine what features in infant death investigations may be significantly associated with medical examiners’ discrimination between SIDS, undetermined, and asphyxia by comparing the number and types of scene risk factors for unsafe sleep-related asphyxia and three additional variables that may affect death certification. These three variables were demonstrable evidence of asphyxia (independent of the number or type of risk factors for asphyxia), presence of a potential competing cause of death, and whether a doll reenactment was done.

METHODS

The NYC OCME is a city-governed agency led by an appointed Chief Medical Examiner who is a board-certified forensic pathologist. The NYC OCME’s jurisdiction covers the five geographic boroughs that comprise the largest city in the United States, with its resident population of just over 8 000 000 people, and averages more than 5000 autopsies per year (19). Death investigations are performed by medicolegal death investigators who are also physician assistants or nurse practitioners. During the study period, the Chief Medical Examiner oversaw an average of 26 full-time forensic pathologists, approximately 90% of whom were board-certified in forensic pathology at any given time in the study period, and an average of four forensic pathology fellows each year. For all but 18 months of the study period, these medical examiners were assigned to one of five physical mortuaries; in the last 18 months, it retained the same geographic jurisdiction and staff but operated three locations.

Researchers conducted a population-based retrospective analysis on sleep-related infant deaths investigated and accepted for jurisdiction by the NYC OCME from January 1, 2005, to December 31, 2015, to describe variables correlated with the certification of death. This time period was searched for all live born infant deaths ≤ 365 days of age. The review was then restricted to those cases in which the infant was last known to be alive when placed to sleep. The causes of death in this study were restricted to SIDS, undetermined, and unsafe sleep-related causes of asphyxia, which included but was not limited to terminology such as positional asphyxia, overlay, entrapment, and wedging. Manner of death was restricted to natural, accident, or undetermined. The review excluded infants not autopsied, infants with an etiologically specific disease found to be the sole cause of a natural death, accidents unrelated to the sleep environment (such as drowning or fires), and infants missing information to evaluate two or more risk factors for asphyxia. Only seven deaths included in the study had one missing variable, which was presumed to be negative and did not affect statistical outcome. The sample resulted in

427 cases of sleep-related infant deaths $\leq$ 365 days old that met the criteria for the study.

Each infant underwent a complete autopsy at the NYC OCME comprised of full-body radiographs, examination of all viscera, histology, and postmortem toxicology. By standard agency protocol, the brains of all infants were examined by a board-certified neuropathologist following formalin fixation and virtually all infants also had postmortem acylcarnitine profile testing. After 2007, nearly all infants had bacterial cultures and virology studies; prior to 2007, virology was not routinely available, though most infants had bacterial cultures done.

Qualitative data was obtained by reviewing the medical examiner case file of all qualified sudden infant deaths. Medicolegal investigation scene reports, scene photographs, autopsy findings, medical records, police reports, and death certificates were reviewed in the case management system of the NYC OCME. The data collected included the assigned case number, age, race, gender, cause of death, and manner of death. Each case was independently reviewed by both a researcher and a forensic pathologist (author MAPS) to assess five known unsafe sleep factors with re-assessment and discussion between the two to resolve any discrepancies.

The scene risk factors for asphyxia were defined as follows:

1. Infant placed to sleep in an unapproved sleep area such as an adult bed, couch, or car seat. Approved infant sleep areas were limited to cribs, bassinets, and playards.
2. Infant positioned on a soft, compressible sleep surface. Firm sleep surfaces were defined as firm mattresses with a fitted sheet and no additional layer of soft bedding between the infant and the mattress. For the purpose of this study, all adult beds were disqualified as firm sleep surfaces.
3. Bed sharing the same sleep surface with other person(s).

4. Soft object(s) or loose bedding in the immediate sleep environment. This was not included as a disqualifying factor for SIDS but was counted as a risk factor for asphyxia in the further analysis of findings that may influence a medical examiner's certification of death.

5. Infant found in prone or side sleep position. Infants found in a prone position on anything other than a firm sleep surface designed for infant safety were disqualified from SIDS when assessing guideline compliance. Prone or side sleep position regardless of sleep surface was counted as a risk factor for asphyxia in the further analysis of findings that may influence a medical examiner's certification of death.

Conformance to intra-agency guidelines was measured by comparing the risk factors for asphyxia with the SIDS-exclusion criteria defined by agency guidelines. By default, all deaths ruled undetermined or asphyxia were in compliance. For deaths ruled SIDS, the presence of any risk factors for asphyxia, as specified by intra-agency guidelines, were found to be non-compliant.

For the second part of the study, the cases were reviewed for documented demonstrable evidence of asphyxia defined as witnessed overlay, entrapment, obstruction of the nose and mouth, or head covered by bedding at the time the infant was found. Evidence of obstruction of the nose and mouth included a description of such by the person finding the infant unresponsive or bilateral symmetric lividity on the face representing a prone and face down position as determined by the forensic pathologist certifying the death.

All autopsy and investigative reports were also reviewed by a forensic pathologist (author MAPS) for diagnoses of natural disease or suspicious factors that were not determined by the medical examiner to be the cause of death but may have been interpreted as a potential competing cause of death if SIDS or asphyxia was otherwise considered. Pathologic findings generally accepted within the medical community as

known to cause, or contribute to, deaths were included as potential competing causes for the purpose of this study. Cases with unresolved suspicions about the behavior of caretakers and/or veracity of the story were also considered to have a competing cause of death since the pathologist may have used that information to lean toward an undetermined cause if a natural or accidental death was otherwise considered.

The cases were reviewed for documentation of a doll reenactment during the scene investigation. Dolls used in reenactments during the study period included featured dolls with movable extremities; weighted, featureless bean bag dolls; and stuffed animals. The use of a doll or stuffed animal of any size used to show the position of the infant when found was included as a reenactment performed.

The infant data was abstracted and then coded for statistical analysis. Descriptive statistics were calculated for categorical variables as percentage and continuous variables as mean, median, and range. The data were subjected to univariate analysis and then multivariate analysis. The significance of the predictors of actual diagnostic certification was assessed by multinomial logistic regression analysis. Odds ratios (OR) and 95% confidence intervals (CIs) were calculated to describe the distribution of responses for all risk factors in the model. All p values were two-tailed and values of $p \leq .05$ were considered significant. Analyses were conducted by using IBM SPSS Statistics software version 20 (IBM SPSS Statistics, Armonk, NY).

General descriptive data obtained on medical examiner personal characteristics were obtained from the NYC OCME personnel department and from personal inquiry for the purpose of providing some context surrounding the medical examiners who certified the deaths. These data included medical examiner age, gender, race/ethnicity, parenthood status, and years of experience practicing full-time forensic pathology. Years of experience and parenthood were based on the person's status at the end of the study period or upon separation from the agency, whichever came first.

RESULTS

Infant Characteristics

Of 427 infant deaths, 249 (58.3%) were male and 178 (41.7%) were female. The average age of death was 82 days. Of the total deaths, 229 (53.6%) were non-Hispanic black infants, 148 (34.7%) were Hispanic, 22 (5.2%) were non-Hispanic whites, 18 (4.2%) were Asian/Pacific Islander, and 10 (2.3%) were other ethnicities. A scene investigation was completed in 424 (99.3%) deaths. Medical examiners ruled 100 (23.4%) deaths as accidental asphyxia due to unsafe sleep, 43 (10.0%) as SIDS, and 284 (66.5%) as undetermined. No death was reclassified as a result of this study. These results are summarized in **Table 1**.

Medical Examiner Characteristics

These infants were examined by a total of 65 medical examiners. Of these, 19 (29.2%) were male and 46 (70.8%) were female, 57 (87.7%) were white, 6 (9.2%) were black, and 2 (3.1%) were Asian. The median age was 40 years (range 30-70) and the median years of forensic pathology experience was 7 years (range 1 to 34). Twenty-six medical examiners (40%) were parents during all or part of the study period and 39 (60%) had no children. Statistical comparisons of personal characteristics in association with certification outcomes could not be performed due to a relatively small number of medical examiners accounting for a disproportionately large number of death certifications, partly due to medical examiner turnover associated with a large fellowship program and some individual predilection for these types of cases; just 16 medical examiners accounted for more than half of the certifications in the study (260/427), skewing the analyses.

Assessment of Conformance to Intra-Agency Guidelines

Medical examiners conformed to intra-agency guidelines when certifying deaths in 406/427 (95.1%) of the deaths. Of the 427 deaths, 75 (17.6%) qualified to be certified as SIDS using the guidelines (**Table 1**), but

only 22 were actually certified by medical examiners as SIDS. Seven deaths that qualified as SIDS were ruled asphyxia and 46 were certified undetermined by medical examiners using their own discretion in evaluating all aspects of the case.

The medical examiners ruled deaths SIDS, despite meeting unsafe sleep exclusion criteria in the guidelines, in 21 deaths (4.9%). Fourteen of these infants met one SIDS-exclusion criterion based on agency guidelines, and seven met two exclusion criteria. When all scene risk factors for asphyxia were considered, eight infants ruled SIDS had one or two risk factors and 13 had three or four scene risk factors for asphyxia. Overall, a total of 13 infants ruled SIDS had demonstrable evidence of asphyxia (**Table 2**).

Compliance was high throughout the study period and was at 100% for four years of the study, including the last two years (**Figure 1**). Five medical examiners had two noncompliant certifications and 11 had one noncompliant certification. No medical examiner had more than two noncompliant certifications.

Assessment of Number and Types of Scene Risk Factors for Asphyxia

Of the 427 deaths, 328 (76.8%) had three or more scene risk factors for asphyxia and 99 (23.2%) had two or fewer risk factors. All five risk factors were found in 110 infants (25.8%). Only six (1.4%) had no risk factor for asphyxia. Of the 328 deaths with three or more risk factors, 13 were ruled SIDS, 89 were ruled asphyxia, and 226 were ruled undetermined. Of the 43 deaths ruled SIDS, 13 had three or four risk factors for asphyxia, 30 infants had one or two risk factors, and 14 infants had just one risk factor (**Table 2**).

Two hundred and ninety decedents (67.9%) were sleeping in an area other than a crib, bassinet, or playard; 339 (79.4%) were on a soft, compressible surface; 233 (54.6%) infants were bed sharing with one or more other persons; and 258 (60.4%) were in a prone or side position (**Table 3**). The most prevalent asphyxia risk factor was the presence of soft objects or loose bedding in the sleep area (377 infants, 88.3%).

Table 1: Demographics and Findings

Characteristics	n (%)
Age of death (days), median (interquartile range)	82 (51-128)
Gender	
Female	178 (41.7)
Male	249 (58.3)
Race/ethnicity	
Asian/Pacific Islander	18 (4.2)
Black, non-Hispanic	229 (53.6)
Hispanic	148 (34.7)
White, non-Hispanic	22 (5.2)
Other	10 (2.3)
Scene investigation	
Done	424 (99.3)
Not done	3 (0.7)
Doll reenactment	
Done	287 (67.2)
Not done	140 (32.8)
Number of SIDS-exclusion criteria by agency guidelines	
0	75 (17.6)
1	65 (15.2)
2	64 (15.0)
3	220 (51.5)
4	3 (0.7)
Demonstrable evidence of asphyxia	
Yes	174 (40.7)
No	217 (50.8)
Unknown	36 (8.4)
Competing cause of death	
Yes	85 (20)
No	342 (80)
Diagnosis allowed by agency guidelines	
Asphyxia or Undetermined	352 (82.4)
SIDS	75 (17.6)
Diagnosis by medical examiner	
Asphyxia	100 (23.4)
SIDS	43 (10.1)
Undetermined	284 (66.5)

SIDS - Sudden infant death syndrome

Soft objects/loose bedding was, by far, the most prevalent risk factor in infants certified as SIDS (38/43 deaths) and was the only risk factor observed in 11 infants ruled SIDS. The most prevalent combination of risk factors was the presence of all five in 110 deaths (25.8%) followed by infants bed sharing in an unapproved bed with a soft surface and loose bedding (92 deaths, 21.5%), prone/side position in a crib/bassinet/playard on a soft sleep surface with loose bedding (43

deaths, 10.1%), prone/side in a crib/bassinet/playard with firm surface but loose bedding (36 deaths, 8.4%), and prone/side but alone in an unapproved bed with a soft sleep surface and loose bedding (34 deaths, 8.0%) (**Tables 3 and 4**)

The presence of three or more risk factors for asphyxia significantly increased the odds of medical examiners certifying either asphyxia (OR 19, $p<0.001$) or

Table 2: Number of Scene Risk Factors for Asphyxia By Death Certification with Other Variables Represented

Doll Reenactment	Demonstrable Evidence of Asphyxia	Competing Cause of Death	Death Certification	Total Number of Scene Risk Factors for Asphyxia						Total	
				0	1	2	3	4	5		
No	No	No	A					2		2	
			S		4	6	2			12	
			U	1	4	3	7	13	10	38	
		Yes	S		1	1				2	
			U	2	5		2	2	1	12	
		Yes	No	A			1	7	8	6	22
				S		2	3		1		6
				U		4	5	5	1	5	20
	Yes		A			2		1	1	4	
			U			2	1	1	3	7	
	Unknown	No	A					1	1	2	
			U				2	7	3	12	
		Yes	U				1			1	
Yes		No	No	A			2		3	1	6
				S		3	5	4			12
	U			2	3	7	9	41	29	91	
	Yes		S		2		1			3	
			U	1	4	2	3	21	8	39	
	Yes		No	A		3	3	16	17	20	59
				S		2	1	1	2		6
				U		1	6	5	12	10	34
		Yes	A				1	1	2	4	
			S				1			1	
			U		1	2	2	4	2	11	
	Unknown	No	A						1	1	
			S					1		1	
			U			3	2	7	6	18	
Yes		U						1	1		
Total				6	39	54	72	146	110	427	

A - Asphyxia, S - Sudden infant death syndrome, U - Undetermined

undetermined (OR 8.9, $p<0.001$) rather than SIDS, whether or not there was demonstrable evidence of asphyxia, a competing cause of death, or a doll reenactment. When comparing medical examiner determination of asphyxia versus undetermined, the odds of certifying a death as asphyxia when there were three or more risk factors for asphyxia was also significant but weaker (OR 2.1, $p=0.04$) (Table 5).

Demonstrable Evidence of Asphyxia (Independent of the Number of Risk Factors for Asphyxia)

In 174 cases (40.7%), demonstrable evidence of asphyxia was present including witnessed overlay, entrapment, evidence of obstruction of the nose/mouth, or head covered by bedding when the infant was found unresponsive. Of these, 13 were ruled SIDS, 89

were ruled asphyxia, and 72 were ruled undetermined. Of the 72 undetermined deaths with demonstrable evidence supporting asphyxia, only 18 (25%) had a potential competing cause of death (Table 2).

Demonstrable evidence of asphyxia significantly increased the odds of medical examiners certifying a death as asphyxia compared to SIDS (OR 24, $p<0.001$) or undetermined (OR 27, $p<0.001$). It did not significantly influence the odds of differentiating undetermined from SIDS (OR 0.9, $p=0.75$) (Table 5).

Potential Competing Causes of Death

A potential competing cause of death was identified in 85 deaths (20%) (Tables 1 and 2). Six infants had documented suspicions about the behavior of caretak-

Figure 1: Compliance with agency guidelines for certification of sudden infant death syndrome (SIDS) deaths.

ers, veracity of the scene story, or autopsy findings of remote injuries noncontributory to the death but suspicious for past abuse. Among competing natural diseases, the most prevalent was evidence of acute or recent respiratory tract infection (27 infants). Premature birth with persistent pulmonary or growth complications was found in nine infants. Various neuropathologic diagnoses were found in 11 infants, developmental or congenital anomalies of unknown significance were documented in six infants, and acute/recent gastroenteritis was found in six infants. A heterogeneous assortment of pathologic findings of questionable or unknown significance affecting all other organ systems accounted for the rest of the potential competing natural diagnoses.

When a potential competing cause of death was present, the odds that a medical examiner would select asphyxia compared to undetermined was significantly less (OR 0.03, $p<0.001$), with or without consideration of other variables. The odds that a medical examiner would certify a death as undetermined compared to SIDS increased in the presence of a potential competing cause of death only after adjusting for all of the other variables (OR 2.9, $p=0.03$). A competing cause of death had no significant bearing on the odds of certifying the cause of death as asphyxia compared to SIDS (OR 0.5, $p=0.28$) (Table 5).

Table 3: Frequency of Individual Asphyxia Risk Factor Types by Certification

Asphyxia Risk Factor Type	Asphyxia	SIDS	UD	Total
A	75	8	207	290
B	91	15	233	339
C	88	38	251	377
D	56	5	172	233
E	73	23	162	258

A - Sleep environment other than crib, bassinet or playard
B - Soft sleep surface
C - Loose bedding or soft object(s) in sleep area
D - Bed sharing with other(s)
E - Prone or side position
SIDS - Sudden infant death syndrome
UD - Undetermined

Doll Reenactment

A doll reenactment was performed in 287 (67.2%) infant deaths (Tables 1 and 2). The presence of a doll reenactment was weakly associated with an increase in the odds of certification of asphyxia or undetermined when compared to SIDS alone (OR 2.0, $p=0.06$; and OR 1.8, $p=0.06$, respectively). However, after adjusting for the other variables (number of risk factors, demonstrable evidence of asphyxia, and

Table 4: Frequency of Asphyxia Risk Factor Combinations by Certification

Combination of Asphyxia Risk Types	Asphyxia	SIDS	UD	Total
None			6	6
A		1	5	6
C	3	11	14	28
E		2	3	5
AB	1	1	2	4
AC	1	2		3
BC	2	2	2	6
BE			3	3
CD		2		2
CE	4	9	23	36
ABC	2	1	12	15
ABD	4		3	7
ABE	3	1	1	5
ACE	1			1
BCE	14	6	23	43
CDE		1		1
ABCD	14		78	92
ABCE	14	2	18	34
ABDE	3		10	13
BCDE	2	2	3	7
ABCDE	32		78	110

None - No known risk factor for asphyxia

A - Sleep environment other than crib, bassinet or playard
B - Soft sleep surface
C - Loose bedding or soft object(s) in sleep area
D - Bed sharing with other(s)
E - Prone or side position
SIDS - Sudden infant death syndrome
UD - Undetermined

competing cause of death), the presence of a doll reenactment failed to show any significant association with the odds of certifying either asphyxia or undetermined compared to SIDS (OR 1.5, $p=0.36$; and OR 1.4, $p=0.4$, respectively). A doll reenactment had no significant bearing on the odds of choosing between asphyxia and undetermined (OR 1.1, $p=0.75$) (**Table 5**).

DISCUSSION

Inconsistencies in investigation and certification of sudden infant deaths are well-recognized (16, 20, 21). Numerous classification systems have been suggested with various ways to describe sudden infant death and attempt to account for unsafe sleep factors that may contribute to death (14, 22). Some classification proposals eliminate the certification of SIDS altogether (23, 24). No means exist at present for establishing or

enforcing a consensus at the national or international levels.

Autopsy pathologists certify deaths largely based on evaluative opinions. Where there is no anatomic injury or disease that is grossly incompatible with life, then the cause of death is more likely to be one entity than others based on the sum total of the findings and the examiner's qualitative estimation of probability. This is inherently more difficult in sudden infant deaths where autopsy and ancillary postmortem laboratory studies most often do not reveal a cause of death and scene information can be difficult to elicit and interpret.

Obstructive or mechanical asphyxia associated with unsafe sleep is a diagnosis of exclusion. There are no anatomic findings or laboratory tests that are diagnostic of asphyxia. Scene investigation findings support-

Table 5: Association Between Case Characteristics and Medical Examiner Certification (n=427)

Case Characteristics	Overall cases, n (%)			Asphyxia vs. SIDS*		Undetermined vs. SIDS*		Asphyxia vs. Undetermined*	
	SIDS 43 (10%)	Asphyxia 100 (23%)	UD 284 (67%)	Univariate Odds Ratio (95% CI), <i>p</i>	Multivariate Odds Ratio (95% CI), <i>p</i>	Univariate Odds Ratio (95% CI), <i>p</i>	Multivariate Odds Ratio (95% CI), <i>p</i>	Univariate Odds Ratio (95% CI), <i>p</i>	Multivariate Odds Ratio (95% CI), <i>p</i>
Number of risk factors for asphyxia									
0-2	30 (7.0)	11 (2.6)	58 (13.6)						
3 or more	13 (3.0)	89 (20.8)	226 (52.9)	19 (7.5-46), <0.001	21 (8-58), <0.001	8.9 (4-18), <0.001	8.7 (4.1-18), <0.001	2.1 (1.0-4.1), 0.04	2.5 (1.1-5.5), 0.03
Demonstrable evidence of asphyxia									
No	29 (6.8)	8 (1.9)	180 (42.2)						
Yes	13 (3.0)	89 (20.8)	72 (16.9)	24 (9-65), <0.001	31 (11-88), <0.001	0.9 (0.4-1.8), 0.75	1.1 (0.5-2.3), 0.83	27 (13-60), <0.001	28 (13-62), <0.001
Unknown	1 (0.2)	3 (0.7)	32 (7.5)	11 (0.9-119), 0.07	6.4 (0.5-74), 0.14	5.1 (0.7-39), 0.11	3.8 (0.5-31), 0.25	2.1 (0.5-8.4), 0.28	1.7 (0.4-7), 0.47
Competing cause of death									
No	37 (8.7)	92 (21.5)	213 (49.9)						
Yes	6 (1.4)	8 (1.9)	71 (16.6)	0.5 (0.2-1.7), 0.28	0.8 (0.2-2.7), 0.68	2.1 (0.8-5.1), 0.12	2.9 (1.1-7.6), 0.03	0.3 (0.1-0.6), 0.001	0.3 (0.1-0.6), 0.003
Doll reenactment									
Not done	20 (4.7)	30 (7.0)	90 (21.1)						
Done	23 (5.4)	70 (16.4)	194 (45.4)	2.0 (0.9-4.2), 0.06	1.5 (0.6-3.6), 0.36	1.8 (0.9-3.6), 0.06	1.4 (0.7-2.7), 0.4	1.1 (0.7-1.8), 0.75	1.1 (0.6-2), 0.75

SIDS - Sudden infant death syndrome

UD - Undetermined

CI - Confidence interval

*Reference group for that comparison

ing asphyxia range from certain (e.g., witnessed overlay), to highly likely (e.g., prone and face down in soft bedding), to inconclusive (e.g., found supine without obstruction of the nose and mouth while bed sharing), to unlikely (e.g., found alone and supine on a bed with nothing around the nose and mouth). Medical examiners and coroners have variably used SIDS, sudden unexplained infant death, asphyxia, and undetermined to describe these infant deaths and have demonstrated differing opinions on the degree of certainty required to determine unsafe sleep-related asphyxia as the cause of death, resulting in inconsistencies in certifications of similar infant deaths across jurisdictions (21). Our study found that infants dying under similar circumstances and with comparable autopsy findings have variable death certifications even within a single agency.

This study showed that internal agency guidelines were successful in one jurisdiction at limiting the number of incorrect SIDS certifications. However, guidelines set forth by Dr. Hirsch offered no guidance in differentiating asphyxia and undetermined, allowing for variable interpretation of deaths with similar findings. The guidelines also fell short of categorically excluding prone/side position from being ruled as SIDS, despite previous studies recognizing that prone/side positions and soft objects/loose bedding are associated with an increased risk of asphyxia and sudden death (10, 11, 18, 25, 26). Of 75 deaths that qualified as SIDS, 69 were in a prone position and/or had loose, soft bedding in the sleep area, which did not disqualify them from SIDS based on intra-agency guidelines. The results suggest that medical examiners are cognizant of these additional risks and self-imposed more stringent criteria than the guidelines; only 22 of the “SIDS-qualified” deaths were certified as SIDS.

This study supported previous conclusions that shared autopsy and similar unsafe sleep findings among most sudden infant deaths are associated with diagnostic overlap among SIDS, asphyxia, and undetermined certifications (8, 27, 28). Many deaths certified as SIDS or undetermined in this study are misclassified based on the presence of unsafe sleep risk factors at the scene and/or demonstrable evidence of asphyxia.

This study found an association between additive risk factors for asphyxia and medical examiners’ exclusion of SIDS, but the number of risk factors for asphyxia are not as strongly associated with differentiation between asphyxia and undetermined. This may be related to individual assessments of risk factors in each case. The perceived contribution of bed sharing, degree of compressibility of a soft sleep surface, and/or threat of the soft objects/loose bedding in the sleep environment are subject to interpretation. The most common scene risk factor in this study was the presence of soft objects or loose bedding in the sleep environment, which is the perhaps the broadest, most vague category subject to interpretation. In reality, in the absence of bed sharing or prone position, it is unlikely that soft bedding or loose objects in the sleep environment not specifically obstructing the airway or covering the head when found bear any physiologic relevance to the infant’s death, despite being objectively identified as a potential risk factor for asphyxia. A tangible number of objective risk factors cannot substitute for professional judgment in assessing individual cases.

The strongest variable associated with certification of asphyxia was demonstrable evidence of asphyxia, even though barely half of infants with this finding were certified as such. In this study, 40.7% of infants had demonstrable evidence of asphyxia, which is slightly less than a previous study that found demonstrable evidence of asphyxia in 51.7% of infant deaths (8). A previous study also found ME/C more likely to certify as asphyxia in deaths with demonstrable evidence of overlay or obstruction of the nose and mouth at the time found unresponsive (21). These studies show relative support among ME/C for a diagnosis of asphyxia when there is demonstrable evidence of asphyxia, suggesting that this would be a good starting recommendation when developing improved guidelines for infant death certification.

The strongest variable associated with a rejection of asphyxia as the cause of death was the presence of a potential competing cause of death. In just six of these deaths, the competing factor was suspicion about the veracity of the scene story, so undetermined may be

most appropriate. In other deaths with a competing factor of natural disease, the possibility of an unsafe sleep-related asphyxia in a medically vulnerable infant should be given due consideration. Acute or recent respiratory tract infection was the most frequent potential competing cause of death in this study and a recent respiratory infection has been previously implicated as a suspected contributing factor in sudden infant deaths, particularly in association with prone sleep (26, 29). The triple risk model outlined by Filiano and Kinney in 1994, proposes that sudden infant deaths may result from the intersection of underlying infant vulnerabilities, a critical developmental period, and an exogenous stressor (30). Research continues to suggest that many sudden infant deaths are multifactorial (26). When the evidence strongly supports asphyxia, it may be relevant to contribute both natural disease and asphyxia associated with unsafe sleep.

A doll reenactment had only a weak possible association with exclusion of SIDS and no statistically significant association with differentiation between asphyxia and undetermined. In a previous survey of ME/C, a doll reenactment was positively reported by more than 60% of respondents to be a “type of evidence” that might cause the certifier to make a determination of an asphyxial death if the reenactment specifically showed demonstrable evidence of asphyxia including obstruction of the nose and mouth, overlay, wedging, or probable neck compression/hanging (21). Of 115 infants in our study who had a doll reenactment and demonstrable evidence of asphyxia, 63 were ruled asphyxia and 45 were ruled undetermined; only 11 of these undetermined deaths had a possible competing cause of death. A reason for reluctance to certify as asphyxia when presented with visual supporting evidence could not be ascertained.

Guidelines could be improved by eliminating SIDS as a certification of infant death, thereby reducing infant death classifications to those caused by etiologically specific natural diseases, unsafe sleep-related asphyxia, and undetermined cause. Guidelines could also be improved by defining situations in which asphyxia should be favored over undetermined as a probable cause, such as deaths in which there is demonstrable

evidence of asphyxia. Undetermined deaths would encompass those in which the cause remains undetermined after a complete investigation but unsafe sleep factors that cannot be ruled out as a contributing factor, those in which the cause remains undetermined after a complete investigation without any known unsafe sleep factors, and those in which the cause remains undetermined because an autopsy or death scene investigation could not be performed (incomplete information). Recognition of these basic categories of sudden infant death and use of corresponding verbiage on the death certificate could then be used to better define International Classification of Disease coding of sudden infant deaths, as proposed by Shapiro-Mendoza et al. (14).

Eliminating SIDS as a certification option would help reduce misclassifications of unsafe sleep-related deaths, but it could limit the ability to separately classify truly natural deaths of unknown etiology that have no reasonable suspicion for asphyxia as a cause or contributing factor. In this study, 30 infants certified as SIDS had just one or two risk factors for asphyxia and 13 of these infants had only objectively identified soft objects or loose bedding in the sleep area and/or were sleeping alone in a supine position on an unapproved surface. These and other similar deaths may have reasonably low probability that asphyxia contributed to the death.

Previous research supports that not all sudden infant deaths are explained exclusively by unsafe sleep factors. Intrinsic factors have been independently associated with increased risk for sudden infant death including but not limited to male gender, premature birth, unspecified/unidentified genetic mutations, and maternal smoking (26). Research has found potential hippocampal and brainstem abnormalities that may be associated with sudden infant death (31, 32). Unsafe sleep does not explain all sudden infant deaths and identifying deaths with low suspicion of asphyxia can help researchers target a subpopulation for further exploration into other causes of sudden infant death.

The study had limitations that should be considered. It was a retrospective analysis in which approximately

25% of the medical examiners certified approximately 60% of the deaths, possibly skewing results in the direction of their certification preferences. The quality of doll reenactments can vary widely but was not assessed in this study and may have some effect on medical examiner interpretation. Beds other than cribs, bassinets, and playards were also counted as soft sleep surfaces because, in theory, a sleep environment outside of an approved infant bed presents its own risk of injury due to entrapment or in combination with other factors and these surfaces (which are most often adult beds or couches) are also commonly known to be soft, compressible surfaces. In reality, these actual risks to the infant of sleeping in an unapproved bed are often one and the same, so the number of risk factors for infants outside of approved beds is inflated by a factor of at least one for many infants in this study.

CONCLUSION

Diagnostic overlap occurs between infants ruled as SIDS, asphyxia, and undetermined, even within a single jurisdiction. Sudden infant death syndrome is a diagnostic distracter where the causes of death are unknown and scene findings are shared by natural deaths of undetermined cause and unsafe sleep-related asphyxias, and it is best eliminated as a certification option. Furthermore, the indiscriminate use of undetermined in deaths with strong supporting evidence of asphyxia limits the capacity of ME/C, epidemiologists, clinicians, and families to recognize and prevent many sudden infant deaths associated with unsafe sleep. Guidelines can be effective in limiting misclassification of deaths, but guidelines for infant death certification need to further define situations that should favor a diagnosis of asphyxia rather than undetermined, such as those with demonstrable evidence of asphyxia or multiple concomitant unsafe sleep factors that make asphyxia more probable than not.

It is cost- and manpower-prohibitive for researchers to review individual case files in depth in jurisdictions across the United States to fully understand the circumstances that drive individual ME/C certifications. Though it may be imperfect, the death certificate is

currently the best tool that ME/C have to communicate factors behind undetermined causes of death for epidemiologic purposes. This can be done using descriptive terminology on the cause of death line (e.g., "Cause undetermined, sudden infant death with unsafe sleep factors" or "Cause undetermined, no autopsy or no death scene investigation") or by utilizing the space in the injury section of undetermined deaths to provide additional information. Where SIDS is discontinued as a certification option, ME/C need a mechanism to convey no reasonable suspicion of asphyxia such as certifying some deaths as undetermined cause with a natural manner and/or adding descriptive information on the death certificate (e.g., "Cause undetermined, no known unsafe sleep factors").

Efforts by ME/C, individually or as a whole, to refine the parameters around types of sudden infant death certification and use the death certificate to better communicate known unsafe sleep factors, or lack thereof, would provide clarity on the circumstances surrounding sudden infant deaths that can help direct research and preventive measures.

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