

# Reclassification of SIDS cases—a need for adjustment of the San Diego classification?

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**Abstract** A study was undertaken reclassifying cases of sudden infant death syndrome (SIDS) taken from two geographically separate locations utilizing the San Diego definition with subclassifications. One hundred twenty-eight infant cases were examined from files at Forensic Science South Australia in Adelaide, SA, Australia over a 7.5-year period from July 1999 to January 2007. Thirty-one cases (24%) had initially been diagnosed as SIDS and 30 (23%) as undetermined while 67 (52%) had an explainable cause of death. After reclassification, the number of SIDS cases had increased to 49 of the 128 cases, now representing 38% of the cases; category IB SIDS constituted 10 (20%) and II SIDS 39 (80%) of the SIDS cases. No cases were classified as IA SIDS. Two hundred eighteen infant cases were identified from the files of the Department of Forensic Medicine, Aarhus University, Denmark over a 16-year period from 1992 to 2007. Eighty-two (38%) were originally diagnosed as SIDS, 128 (59%) with identifiable causes of death, and 8 (4%) as unexplained. After review, 77 (35%) cases were reclassified as SIDS, a decrease of 6%. Twenty (26%) infants were classified as category IB SIDS and 57 (74%) as II SIDS. None of the cases met the criteria for IA SIDS. Problems arose in assessing

cases with failure to thrive, fever, and possible asphyxia. Modifications to the San Diego subclassifications might improve the consistency of categorizing these cases.

**Keywords** Sudden infant death syndrome (SIDS) · IB SIDS · II SIDS · Unexplained sudden infant death (USID)

## Introduction

The San Diego definition of sudden infant death syndrome (SIDS) was formulated at a meeting held in San Diego in 2004 [1]. The objectives of the San Diego definition were to address some of the weaknesses of previous definitions, which were strictly exclusive and often did not include positive criteria such as an age range and the occurrence of death during sleep [1]. The term sudden infant death syndrome was defined in 1969 as “the sudden death of any infant or young child which is unexpected by history, and in which a thorough post-mortem examination fails to demonstrate an adequate cause of death” [2]. Many definitions and criteria have since been proposed but only a small number have had international acceptance. Unfortunately interpretations of SIDS definitions have been numerous, inconsistent, and idiosyncratic [3–11]. Autopsy and investigative protocols have also varied, and in some western countries, death can be attributed to SIDS even though autopsies have not been performed in more than 40% of cases [12]. As a consequence, comparisons among different countries or even between institutions in the same country should never be carried out without ensuring that a uniform classification of SIDS cases has been followed, or preferably only if cases have been reclassified according to uniform criteria. Currently these issues make the assessments of the true differences in SIDS incidence between

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Western countries difficult, with even more problems with comparisons involving the rest of the world [13]. This is a major deficiency in SIDS research.

The purpose of this study was to investigate the usefulness of the San Diego definition in reclassifying cases and to identify the types of cases that caused the most difficulties with reclassification. Furthermore, the reasons for classifying and subclassifying sudden infant cases according to the San Diego criteria were examined in two separate populations: in South Australia, Australia, and in the northern part of Jutland, Denmark.

## Materials and method

Case files from Forensic Science South Australia (FSSA) in Adelaide, SA, Australia were examined for all sudden infant deaths over a 7.5-year period from July 1999 to January 2007, together with case files from the Department of Forensic Medicine, Aarhus University, Denmark over a 16-year period, from 1992 to 2007. All cases had undergone full police investigations and full autopsy examinations. In addition, death scene and coronial investigations had been performed for all of the Australian cases. FSSA serves a population of approximately 1.6 million and the Department of Forensic Medicine at Aarhus University serves a population of approximately 2.2 million. Details of the cases were reviewed, including the circumstances of deaths, the available medical history of the infants, the causes and manners of the deaths, and any significant autopsy findings, histology results, and the results of ancillary tests. The cases were subsequently reclassified according to the San Diego criteria as either IA SIDS, IB SIDS, II SIDS, unexplained sudden infant death (USID), or explained [1] (Fig. 1). Reasons for reclassifying each case were noted.

In the South Australian material, the cases had initially been classified using Beckwith's original definition, with the NICHD definition being used after 1991. After 1992, the autopsies had been performed according to the Australasian Autopsy protocol [2, 3, 14]. All Danish cases from 1992 had originally been classified according to the Nordic SIDS criteria that had been developed by the Nordic pathology cooperation NORD PAT in 1990 and launched at the Third International Conference on SIDS in Stavanger, Norway, 1994 [15]. According to this definition, sudden infant death cases were divided into three categories:

1. Pure SIDS cases in which the autopsy and clinical information do not reveal any cause of death
2. Borderline SIDS cases, in which preexisting congenital disorders or clinical symptoms, and/or post-mortem findings, are not severe enough to explain the death.

3. Non-SIDS cases, in which the cause of death is explained according to clinical information and/or the results of the post-mortem examination [7].

All Danish cases had been coded by a pathologist using a system based on the SNOMED system in which the code “unexplained infant death” is equivalent to “SIDS”. Once a year, all cases had been reviewed by the same senior pathologist to ensure consistency in the way the cases were being coded.

## Results

### Australian cases

#### *Results of the reclassification*

In the Australian material, 128 infant cases were identified. Thirty-one cases (24%) had initially been diagnosed as SIDS and 30 (23%) as undetermined while 67 (52%) had an explainable cause of death. After reclassification, the number of SIDS cases had increased to 49 of the 128 cases, now representing 38% of the cases; category IB SIDS constituted 10 (20%) and II SIDS 39 (80%) of the SIDS cases. No cases were classified as IA SIDS. There were now 26 undetermined cases (20%) and 53 cases with an explained cause of death (41%). The number of IB SIDS was constant at around 0–2 cases per year, category II SIDS varied between 3 and 8 cases, USID between 1 and 6 cases, and “explained” between 4 and 11 cases (Fig. 2).

In the IB SIDS category, nine (90%) had originally been classified as SIDS and one (10%) as undetermined (Table 1). Among the II SIDS cases, 22 (56%) had previously been categorized as SIDS, 14 (36%) as undetermined, and 1 (3%) as overlaying. Two (5%) postresuscitation cases had met the criteria for II SIDS. There were no cases originally diagnosed as SIDS in the USID or “explained” categories.

#### *Reasons for the subclassification*

The reason that no cases met the requirements for category IA SIDS was that there was insufficient ancillary testing, primarily metabolic analysis which was absent in 69% of all cases. The reasons for classifying cases as category II, and not IB SIDS were most commonly a lack of information about neonatal and perinatal conditions, for example those resulting from preterm birth which was lacking in 17 (27%) of the cases (Table 3). The second most common cause was that mechanical asphyxia or suffocation had not been ruled out with certainty in 13 (20%) of the cases. Infants born preterm or with other perinatal conditions now resolved was the third most common reason for

**Fig. 1** The San Diego criteria [1]

<p><b>General Definition of SIDS</b></p> <p>SIDS is defined as the sudden unexpected death of an infant &lt;1 year of age, with onset of the fatal episode apparently occurring during sleep, which remains unexplained after a thorough investigation, including performance of a complete autopsy and review of the circumstances of death and the clinical history.</p> <p><b>Category IA SIDS: Classic Features of SIDS Present and Completely Documented</b></p> <p>Category IA includes infant deaths that meet the requirements of the general definition and also all of the following requirements.</p> <p><i>Clinical</i></p> <ul style="list-style-type: none"> <li>• More than 21 days and &lt;9 months of age.</li> <li>• Normal clinical history, including term pregnancy (gestational age of <math>\geq 37</math> weeks).</li> <li>• Normal growth and development.</li> <li>• No similar deaths among siblings, close genetic relatives (uncles, aunts, or first-degree cousins), or other infants in the custody of the same caregiver.</li> </ul> <p><i>Circumstances of Death</i></p> <ul style="list-style-type: none"> <li>• Investigation of the various scenes where incidents leading to death might have occurred and determination that they do not provide an explanation for the death.</li> <li>• Found in a safe sleeping environment, with no evidence of accidental death.</li> </ul> <p><i>Autopsy</i></p> <ul style="list-style-type: none"> <li>• Absence of potentially fatal pathologic findings. Minor respiratory system inflammatory infiltrates are acceptable; intrathoracic petechial hemorrhage is a supportive but not obligatory or diagnostic finding.</li> <li>• No evidence of unexplained trauma, abuse, neglect, or unintentional injury.</li> <li>• No evidence of substantial thymic stress effect (thymic weight of &gt;15 g and/or moderate/severe cortical lymphocyte depletion). Occasional “starry sky” macrophages or minor cortical depletion is acceptable.</li> <li>• Negative results of toxicologic, microbiologic, radiologic, vitreous chemistry, and metabolic screening studies.</li> </ul> <p><b>Category IB SIDS: Classic Features of SIDS Present but Incompletely Documented</b></p> <p>Category IB includes infant deaths that meet the requirements of the general definition and also meet all of the criteria for category IA except that investigation of the various scenes where incidents leading to death might have occurred was not performed and/or <math>\geq 1</math> of the following analyses was not performed: toxicologic, microbiologic, radiologic, vitreous chemistry, or metabolic screening studies.</p> <p><b>Category II SIDS</b></p> <p>Category II includes infant deaths that meet category I criteria except for <math>\geq 1</math> of the following.</p> <p><i>Clinical</i></p> <ul style="list-style-type: none"> <li>• Age range outside that of category IA or IB (i.e., 0–21 days or 270 days [9 months] through first birthday).</li> <li>• Similar deaths among siblings, close relatives, or other infants in the custody of the same caregiver that are not considered suspect for infanticide or recognized genetic disorders.</li> <li>• Neonatal or perinatal conditions (for example, those resulting from preterm birth) that have resolved by the time of death.</li> </ul> <p><i>Circumstances of Death</i></p> <ul style="list-style-type: none"> <li>• Mechanical asphyxia or suffocation caused by overlaying not determined with certainty.</li> </ul> <p><i>Autopsy</i></p> <ul style="list-style-type: none"> <li>• Abnormal growth and development not thought to have contributed to death.</li> <li>• Marked inflammatory changes or abnormalities not sufficient to be unequivocal causes of death.</li> </ul> <p><b>Unclassified Sudden Infant Death</b></p> <p>The unclassified category includes deaths that do not meet the criteria for category I or II SIDS but for which alternative diagnoses of natural or unnatural conditions are equivocal, including cases for which autopsies were not performed.</p>
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classifying a SIDS case as category II SIDS, involving ten (16%) of the cases (Table 3).

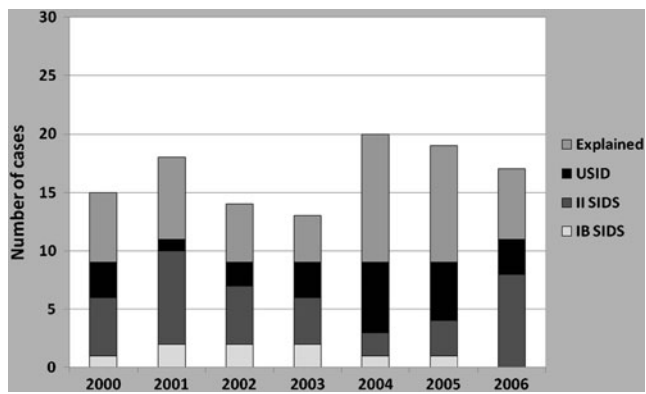
Danish cases

#### Results of the reclassification

A total of 218 infant cases were identified of which 82 (38%) were originally diagnosed as SIDS, 128 (59%) with identifiable causes of death, and 8 (4%) as unexplained. After review, 77 (35%) cases were reclassified as SIDS according to the San Diego definition, a decrease of 6%. Twenty (26%) infants

were classified as category IB SIDS and 57 (74%) as II SIDS. None of the cases met the criteria for IA SIDS as vitreous humor screening was not (and still is not) a part of the Danish SIDS protocol. In addition, death scene investigation had not been performed in seven of the IB SIDS cases.

From 1992 to 2007, the number of SIDS cases has declined from 14 to approximately 2 SIDS cases per year, with the decrease being most prominent for the IB SIDS category. For example, no cases were classified as IB SIDS over the past 6 years (2002–2007) of the study. The number of cases classified as “explained” was relatively stable with a mean value at 7.2 cases per year (2–12) (Fig. 3).



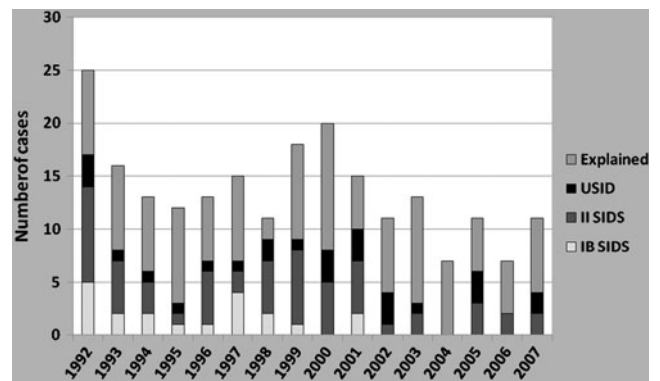
**Fig. 2** Distribution of Australian infant cases classified according to the San Diego criteria over time (2000–2006)

All the category IB SIDS cases and 51 (89%) of the II SIDS cases had originally been classified as SIDS (Table 2). Of the two remaining category II SIDS cases, one had significant cerebral edema with no identifiable cause, and the second had been classified as “unexplained” with an upper airway infection.

Of the ten SIDS cases that were reclassified as USID: four had unexplained injuries, one had a sibling death with insufficient information on the death, one had a perinatal condition that had not resolved by the time of death, two had failure to thrive with signs of dehydration, one had a rectal temperature of 42°C measured during attempted resuscitation, and one had achondroplasia and had been under investigation at hospital for previous episodes of

**Table 1** Results of reclassification using the San Diego criteria compared to the original diagnoses from autopsy reports—Australian cases

Australian cases	
Original diagnoses	Number of cases (%)
SIDS 1B	
SIDS	9 (90)
Undetermined	1 (10)
SIDS II	
SIDS	22 (56)
Undetermined	14 (36)
Postresuscitation cases	2 (5)
Possible overlaying	1 (3)
USID	
Undetermined	10 (38)
Undetermined consistent with explained cause	3 (12)
Various explained caused	13 (50)
Explained	
Undetermined consistent with failure to thrive	2 (4)
Various explained causes	51 (96)



**Fig. 3** Distribution of Danish infant cases classified according to the San Diego criteria over time (1992–2007)

apnea and cyanosis. Achondroplasia is known to increase the risk of sudden death in infants due to foramen magnum stenosis [16]. In the current case, the original case files were missing and with no information on whether the foramen magnum in fact had stenosis; it was decided to classify the case as USID instead of explained. One of the cases originally classified as SIDS had evidence of overlaying/asphyxia with numerous petechiae on the thorax, face, and neck and was reclassified as an explained death.

#### Reasons for the subclassification

In the Danish material, the main reasons for reclassifying a category IB SIDS case to category II SIDS were that mechanical asphyxia had not been ruled out (23 cases (21%)) and that inflammatory changes/abnormalities had been found that were not considered to be sufficient to cause death (21 cases (19%)). Missing information concerning birth and whether the child was born preterm accounted for the third leading cause in 20 (18%) of the cases (Table 3).

#### Discussion

A limitation of this study is that case reviews were restricted to material that was present in files in the respective forensic institutes. In addition a certain number of histology reports were insufficiently detailed for the authors to be able to determine with certainty whether the case should be classified as SIDS or an explained death [17]. This was particularly so in the Danish cases, as 30 (14%) of the Danish infants had pneumonia as the cause of death, while this was only the case in 2 (2%) of the Australian cases. Infants with lung infections or minor inflammatory infiltrates are known to cause classification problems [18] and this has previously been reported from other studies using Nordic material [19].

**Table 2** Results of reclassification using the San Diego criteria compared to the original diagnoses from autopsy reports—Danish cases

Danish cases	
Original diagnoses	Number of cases (%)
IB SIDS	
SIDS	20 (100)
II SIDS	
SIDS	51 (89)
Brain edema, cause unknown	1 (2)
Interstitial pneumonia and/or upper airway infection	5 (9)
USID	
SIDS	10 (40)
Undetermined	8 (31)
Various explained causes	8 (31)
Explained	
SIDS	1 (1)
Various explained causes	115 (99)

One of the most notable differences between the Australian and the Danish material was the alteration in percentage of SIDS cases after review. In the Australian material, the number of SIDS cases increased by 58% (31 to 49 cases) while a decrease of 6% occurred in the Danish cases (82 to 77 cases). This marked difference could be due to the use of different SIDS definitions (or possibly to differences in the interpretation of findings) and only serves to emphasize the importance of using identical definitions

when comparing SIDS data. The change in the Australian data could also be due to an increasing trend in recent years for pathologists to avoid the term SIDS in favor of “undetermined”. Marked variations in SIDS numbers and SIDS subclassifications have been reported previously and are a well-recognized problem in the field [20, 21]. Even with similar SIDS definitions, significant differences are reported, although studies with high concordance among pathologists have been reported [15, 20]. With the decline SIDS cases, it would be appropriate for pathologists to select standard definitions, to use established protocols (including death scene examinations), and to also participate in multidisciplinary discussions where significant clinical and other information will be available to enable the most appropriate classifications to be made [13, 22–24].

The low number of Danish cases initially diagnosed as undetermined compared to the Australian is probably a result of two factors in combination. First of all the different classification criteria are in themselves expected to cause some differences in the number of cases diagnosed as undetermined, and secondly all Danish cases had been reviewed by the same pathologist, which furthermore could have lowered the number of undetermined cases.

No category IA SIDS cases were found in either the Australian or Danish material primarily because of insufficient ancillary testing. This was expected, as the archival material was collected using protocols that did not require ancillary tests necessary for classifying cases as IA SIDS. For this reason, it has been suggested that the IA SIDS category should be merged with IB [25]. However, we find

**Table 3** Reasons for subclassifying SIDS cases as either IB SIDS or II SIDS in the Australian and Danish infant cases; there were no cases classified as IA SIDS

	AUS <i>n</i> (%)	DK <i>n</i> (%)
IB SIDS instead of IA SIDS		
Insufficient ancillary testing	10 (16)	20 (18)
No death scene investigation	0 (0)	7 (6)
II SIDS instead of IB SIDS	Two reasons in 11 cases; 3 reasons in 2 cases	Two reasons in 22 cases; 3 reasons in 3 cases
Lack of information concerning birth (term/preterm?)	17 (27)	20 (18)
Mechanical asphyxia not ruled out	13 (20)	23 (21)
Preterm/perinatal conditions now resolved	10 (16)	12 (11)
Inflammatory changes/ abnormalities	5 (8)	21 (19)
Abnormal growth/development not thought to contribute to death	4 (6)	3 (3)
Age outside range (0–21 or 270–365 days)	2 (3)	3 (3)
Other	2 (3)	2 (2)
Similar deaths among siblings	1 (2)	1 (1)
Total	64 (100)	112 (100)



it a useful distinction to enable distinguishing cases with scene investigation and extensive testing from those that are missing important information.

Information that was most often lacking concerned whether an infant had been born preterm, and this unfortunately proved to be a major problem in both the Australian and Danish data resulting in cases moving from categories IB to II SIDS. We suspect that the majority of these infant were, in fact, born at term and that this information was merely underreported by parents, police, general practitioners, and pathologists.

Even when pathologists use the same definition of SIDS, certain cases will be open to interpretation and disagreement. The greatest degree of disagreement in classification occurred in cases of infants with failure to thrive, fevers, or possible asphyxia.

One of the major differences between previous definitions and the San Diego definition is the exclusion of cases with evidence of unexplained trauma, abuse, or neglect. In the Danish cases, this resulted in the reclassification of four cases from SIDS to USID. A review of cases from the German SIDS Study (GeSID) using the San Diego definition also showed a decrease in cases classified as SIDS, and the exclusion of four cases due to evidence of trauma or a suspicion of neglect [25]. The advantage of including these exclusion criteria is that it is less likely that homicides will be misclassified as SIDS when children with unexplained injuries are being excluded as it has been clearly shown that infants with bruises and other nonlethal injuries have a significantly higher risk of death due to homicide [26]. However, it is notoriously difficult to prove neglect as it is characterized by omission of behavior [27]. Some clarification of when “abnormal growth not thought to contribute to death” becomes “evidence of neglect” is probably needed; this could include factors such as infant weight below the third percentile against a background of parental factors such as substance abuse, mental impairment, and psychiatric illness etc.

In the review of the Danish cases, three SIDS cases with elevated rectal temperatures recorded during resuscitation attempts or at post-mortem were identified. One infant with a temperature of 42°C was reclassified as USID, given the degree of the fever. Two other cases had recorded rectal temperatures of 40.5°C and 40.8°C with negative histories, bacteriology, and histology. Both infants were appropriately wrapped for the ambient temperatures and so were classified as category II SIDS, as the elevated temperatures in isolation were not deemed to be enough to be fatal. Furthermore in the absence of guidance from the San Diego definition, the GeSID criteria classifies cases with rectal temperatures more than 40°C but less than 42°C as SIDS with more severe findings (SIDS+) [25]. Even though it can be argued that a temperature of 40°C is not fatal in itself, it is highly suggestive of infection especially in the absence of

other causes of hyperthermia. A recent study of children 2 to 36 months of age with fever  $\geq 39$ , a pathogen was only confirmed in 14% of the cases [28]. For that reason, evidence of elevated temperature during resuscitation is just a valid reason for degrading a case from IB SIDS to II SIDS as inflammatory findings are not sufficient to be unequivocal causes of death and adding temperature criteria to the San Diego definition might be a simple and very helpful adjunct.

According to the San Diego criteria, cases where “mechanical asphyxia or suffocation caused by overlaying” is not determined with certainty are classified as category II SIDS. Cases where other causes for mechanical asphyxia such as covering of the mouth or where positional asphyxia is suspected are not addressed in the San Diego classification. In the current study, it was decided to classify these cases as category II SIDS as IB requires that the infant was found in a “safe sleeping environment with no evidence of accidental death”. However, it could be argued that these cases should be classified as USID. Given the difficulties that arise in the diagnosis of asphyxia in the young [11], clarification of this in a revision of the definition would be useful.

## Conclusion

A reclassification of infant cases according to the San Diego definition resulted in an increased number of SIDS cases in data from Australia and a decreased number in the Danish material. The general San Diego definition was found to be practical and the subclassifications provide a guide to the level of investigation that had been undertaken and the certainty to which the diagnosis of unexpected infant death had been made; however, an adjustment addressing some frequently arising issues such as failure to thrive, fever, and suspected asphyxia would enhance the classification and improve comparability between SIDS data from different sources.

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