

## Study and prevention of sudden infant death: An experience in the Valencian community

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### ARTICLE INFORMATION

Received: April 18, 2017

Accepted: May 18, 2017

### Competing interests

The authors declare no competing interests

### Acronyms

**SD:** sudden death

**SIDS:** sudden infant death syndrome

On-Line Versions:

Spanish - English

### ABSTRACT

**Introduction:** The sudden infant death syndrome (SIDS) is the first cause of death in the first year of life. In addition to the of San Diego definition, the autopsy must be complemented with clinical data and examination of the circumstances of death.

**Objective:** To apply this classification in cases of sudden infant death.

**Method:** Multidisciplinary study in 109 infants who died suddenly between September/2006 and December/2015 and who were admitted to the Institutes of Legal Medicine of the Valencian community in Spain. An examination of the involved place, collection of perinatal data, epidemiological survey and judicial autopsy with complementary studies were carried out.

**Results:** From the 109 cases, 60 males and 49 females, 53 (48.6%) were diagnosed as SIDS: 6 IB category, 47 II category (36 with risk factor of accidental suffocation with 19 co-sleeping, 13 prone and 1 with both), 46 cases (42.2%) were due to an explained cause of death (54.4% by infectious diseases, 30.4% heart diseases, 2.2% neonatal hemochromatosis and immunodeficiency, 4.3% congenital malformations and, 2.2 % violent death) and 10 cases (9.2%) of undetermined cause. The average age at death was of 3.53 months. The higher incidence of co-sleeping in SIDS took place at 2 months, of prone position to 4, and most cases presented during the winter.

**Conclusions:** The multidisciplinary study of sudden infant death provides key information for classifying it by categories and for confirming the presence of risk factors, enabling to focus the prevention measures and the family cardiological study.

**Key words:** Sudden infant death, San Diego definition, Risk Factors, Co-sleeping

### *Estudio y prevención de la muerte súbita infantil: Experiencia en la comunidad valenciana*

### RESUMEN

**Introducción:** El síndrome de muerte súbita del lactante (SMSL) es la primera causa de muerte en el primer año de vida. Además de la clasificación de San Diego, la

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*autopsia debe complementarse con datos clínicos y examen de las circunstancias de la muerte.*

**Objetivo:** *Aplicar esta clasificación en casos de muerte súbita de lactantes.*

**Método:** *Estudio multidisciplinar en 109 lactantes que fallecieron de forma súbita entre septiembre/2006 y diciembre/2015, y que ingresaron en los Institutos de Medicina Legal de la comunidad valenciana, en España. Se realizó examen del lugar del levantamiento, recogida de datos perinatales, encuesta epidemiológica y autopsia judicial con estudios complementarios.*

**Resultados:** *De los 109 casos, 60 masculinos y 49 féminas, 53 (48,6%) fueron diagnosticados como SMSL: 6 categoría IB, 47 categoría II (36 con factor de riesgo de sofocación accidental con 19 colechos, 13 pronos y 1 con ambos), 46 casos (42,2%) fueron muerte de causa explicada (54,4% enfermedades infecciosas, 30,4% enfermedades cardíacas, 2,2% hemocromatosis neonatal e inmunodeficiencia, 4,3% malformaciones congénitas y 2,2% muerte violenta) y 10 casos (9,2%) de causa indeterminada. La edad media al fallecimiento fue de 3,53 meses. La mayor incidencia de colecho en el SMSL ocurrió a los 2 meses, de decúbito prono a los 4, y el mayor número de casos se presentó en invierno.*

**Conclusiones:** *El estudio multidisciplinar de la muerte súbita del lactante proporciona información clave para clasificarlas por categorías y confirmar la presencia de factores de riesgo que permitan dirigir las medidas de prevención y el estudio cardiológico familiar.*

**Palabras clave:** *Muerte súbita del lactante, Clasificación de San Diego, Factores de riesgo, Colecho*

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## INTRODUCTION

There is no universal definition for sudden death (SD), however it is generally defined as a natural death which occurs unexpectedly in a healthy person with unknown serious illness in a short time period from symptom onset (generally within 1 hour of symptom onset and death); but different intervals (2, 6 and 24 hours) have been proposed for specific circumstances such as unwitnessed death. Nearly 12.5% of natural deaths are estimated to be sudden deaths (2 hours after the onset of symptoms are considered to be the limit)<sup>1</sup>.

According to this definition, sudden infant death occurs in children under 14 years of age and includes two periods: the first year of life, except the first week (perinatal death) and from the first year to 14 years of age<sup>2</sup>. Only a small number of natural deaths in children are sudden and unexpected; percentages range from 2.3-8% to 20%<sup>2</sup>.

The most frequent causes of sudden infant death are similar in both groups, apart from sudden infant death syndrome (SIDS) in the first year, and include infections and congenital anomalies. Concepts, causes and their classification are widely discussed in the **supplementary material**<sup>3-8</sup>.

This type of case requires every investigation protocol to be standardized: parental interview, corpse removal and autopsy; which, unavoidably, leads to create multidisciplinary working teams. Thus, excellence addressing sudden death in general, and sudden infant death in particular, should bring together pathologists, forensics, cardiologists, pediatricians, microbiologists, epidemiologists, psychologists, among others in the same scope of work. The need for a multidisciplinary-multiagency effort to address SIDS led to the creation in Valencia, in 2006, of a “*Grupo de Trabajo Multidisciplinar de SMSL*” (SIDS Multidisciplinary Working Team) formed by all of the aforementioned specialists from the three provinces of the Valencian Community, whose objectives coincided with those of the “*Asociación Española de Pediatría (AEP)*” (Spanish Association of Pediatrics)<sup>3</sup>. They joined the “*Unidad de Valoración del Riesgo de Muerte Súbita Familiar (UVRMSF)*” (Family Sudden Death Risk-Assessment Unit) in March 2008, where, in addition to SIDS cases and seemingly lethal episodes, they study from a multidisciplinary perspective, the families of victims (deceased or resuscitated) due to potentially familial sudden cardiac death.

The objective of the research has been to apply the San Diego classification in the cases studied.

**METHOD**

A multidisciplinary study was carried out applying the San Diego classification to a sample of 109 apparently healthy-infants under one year of age (from birth to 12 months of age) who died suddenly and unexpectedly and were referred to the Institutes of Legal Medicine and Forensic Sciences in the three provinces of the Valencian Community (Castellón, Valencia, Alicante), in Spain, for medico-legal autopsy, between September 2006 and December 2015.

The study involved different professionals (pediatricians, cardiologists, forensic doctors, pathologists and psychologists), with the following action protocol: death scene investigation and circumstances of death, perinatal data collection, epidemiological survey, medico-legal autopsy with complementary studies (histopathological, toxicological, biochemical, microbiological and metabolic), cardiological study (channelopathy screening) to relatives of cases still classified as SIDS and, if there were siblings at risk, referral to the pediatrician for home monitoring.

When complementary study results were obtained, cases were categorized and closed, and statistical data-processing was done through Power BI program.

**RESULTS**

Over a 9-year period of study, an average 11 cases were obtained per year, with a higher incidence in 2008, 16 in total (14.7%), and only 4 (3.7%) in 2015 (Figure 1). Despite a decreasing trend in the number of cases, SIDS proportion remains consistent every year for nearly 50%.

General average age at death is 3.53 months, and 4.24 months when there is an explained cause. In the case of SIDS, the average age is 3.13 months, and in cases from unexplained cause, 2.40 months. By age groups, the highest number of cases (53 deaths) was registered between the first and third month of life, representing a 48,6% (Figure 2).

Table 1 shows the distribution of the deceased according to sex; with male predominance in all categories.

When applying the San Diego classification, 46 (42,2%) out of the 109 cases studied, had an explain-

able cause of death. In 10 (9.2%) the cause remained undetermined while 53 (48,6%) were diagnosed as SIDS. In the latter, no cases were classified as SIDS IA, but 6 were classified as SIDS IB and 47 as category II, 36 of them presented some risk factor for accidental suffocation (Figure 3): co-sleeping (52.8%), prone position (36.1%) and both (2.8%). These category II cases with a risk factor for accidental suffocation represented 33% of the total cases and 67.9% of those diagnosed as SIDS.

Concerning risk factors for accidental suffocation and the average age, the highest incidence of co-sleeping occurs at 2 months while the highest incidence of prone position at 4 months.

Table 2 details the diagnosis of the 46 ex-

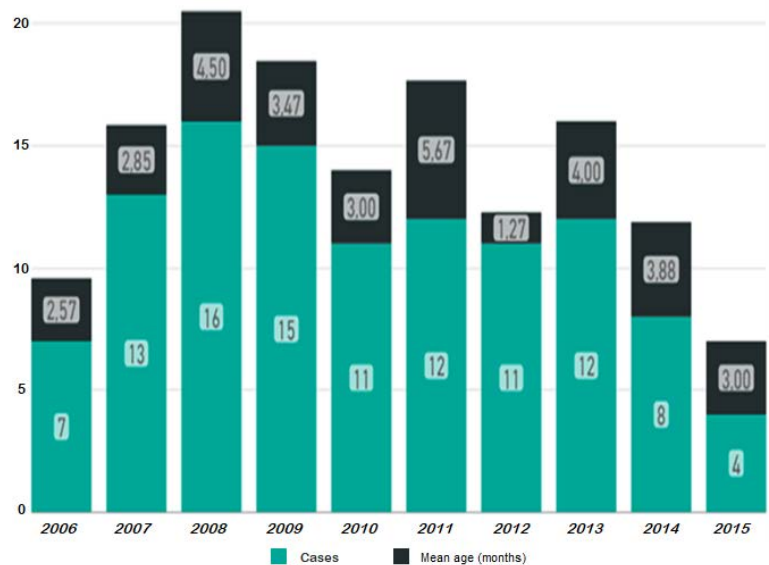


Figure 1. Annual and average distribution of the deceased's age.

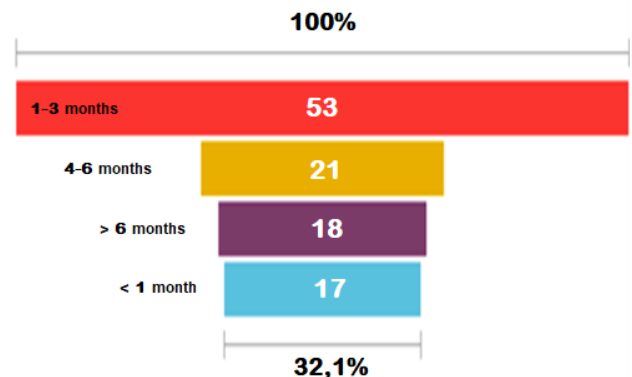
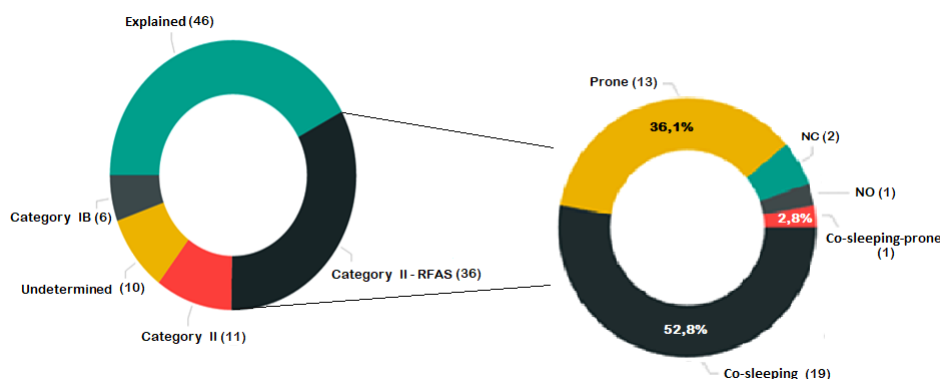


Figure 2. Age groups at the time of death.

**Table 1.** Distribution of the deceased according to sex.

Category (Nº of cases)	Sex			
	Female		Male	
	Nº	%	Nº	%
Total cases (109)	49	44,9	60	55,1
SIDS (53)	23	43,4	30	56,6
Death from explained cause (46)	19	41,3	27	58,7
Death from unexplained cause (10)	3	30,0	7	70,0

SIDS, sudden infant death syndrome.



**Figure 3.** Distribution of cases according to the categories of the San Diego classification (left) and according to the risk factors for accidental suffocation (RFAS) (right).

**Table 2.** Explained causes of death.

Diagnosis	Nº	%
<b>Infectious diseases</b>	<b>25</b>	<b>54,3</b>
- Sepsis	11	23,9
- Pneumonia-Bronchopneumonia-Bronchiolitis	10	21,7
- Meningitis	3	6,5
- Myocarditis	1	2,2
<b>Heart diseases</b>	<b>14</b>	<b>30,4</b>
- Congenital heart disease	7	15,2
- Channelopathies	3	6,5
- Cardiomyopathies	4	8,7
<b>Immunodeficiency</b>	<b>1</b>	<b>2,2</b>
<b>Congenital malformations</b>	<b>2</b>	<b>4,3</b>
<b>Hemochromatosis</b>	<b>1</b>	<b>2,2</b>
<b>Violent death</b>	<b>1</b>	<b>2,2</b>
<b>Congenital valleculea cyst/ Alteration of the cardiac CS</b>	<b>1</b>	<b>2,2</b>
<b>Hypoxic-ischemic brain injury- accidental suffocation</b>	<b>1</b>	<b>2,2</b>
Total	46	100

CS, conduction system

explained death cases, representing 42.2% of the total.

In these deaths, the highest average age was 8 months in cases of bronchopneumonia/ bronchiolitis, and the lowest was 1 month in cases of congenital diseases.

Valencia, the province with the largest population with an estimated 2.5 million inhabitants, reported the highest number of cases (61.5%), followed by the province of Alicante (27.5%), with less than half of cases despite having a population difference of almost 600 thousand inhabitants less than Valencia; and Castellón, with nearly 600 thousand inhabitants, is the province that reported the fewest cases (11%). However, in the three provinces, infants had a similar average age at death.

No seasonal rhythm is seen in the distribution by

months. The highest incidence of cases was registered in January (18 cases, 16.5%), followed by February and November 12 cases each (11.01%), October 11 (10.01%) and June 10 (9.17%). Months with the lowest number of cases were May and August, 4 (3.7%) in each month.

Of the 53 SIDS cases, January and February (9 cases in each month [17%]), followed by November with 7 (13.2%), were the months with highest incidence. April (1 case, 1.9%), July and August, 2 each (3.8%), had the lowest incidence.

In the 46 cases of death from explained causes, January was the month with highest incidence, 9 cases (19.6%), followed by October, 6 (13%) and April, 5 (11%). March and August had the lowest incidence, 2 cases (4.3%) in each month, and May with 1 (2.2%).

Winter and fall presented the highest incidence of occurrence, with 37 (34%) and 31 (28.44%), respectively; 21 (19.26%) in spring and 20 (18.34%) in summer. **Figure 4** shows the distribution (according to each season of the year) of SIDS, in cases with explained/unexplained death and of the de-

ceased's average age.

## DISCUSSION

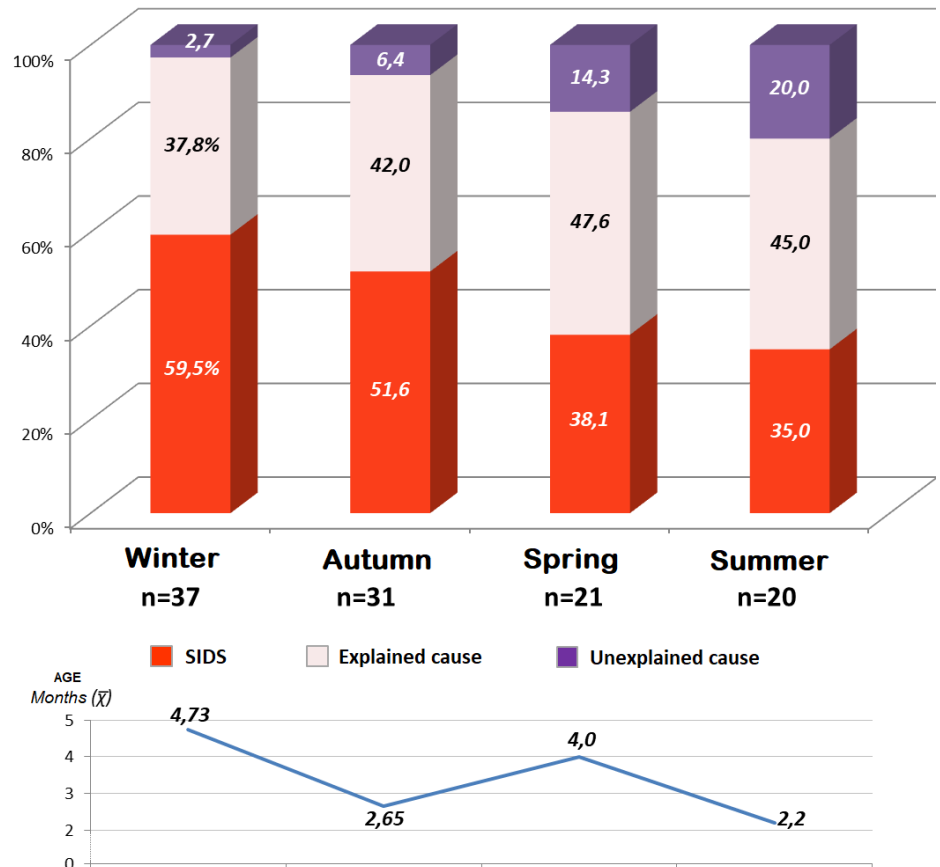
There is a high percentage of SIDS cases associated with unsafe sleep environment (68%). In this series they are included in category II of San Diego, since, according to the survey results and post mortem examination, there is a suspicion of possible asphyxia or suffocation due to "unsafe sleep environment", such as prone positioning, co-sleeping, or overlaying, not determined with certainty. Some schools<sup>2-5</sup> hold that the intervention of a possible external factor in the cause of death is critical to rule out SIDS and diagnose SUID-Sudden Unexpected Infant Death, a term to describe the sudden death of a baby less than 1 year old where autopsy does not reveal a clear cause of death or circumstances suggest some external factor, failing to determine how much contributed to death".

Regardless of their designation, it seems clear that these cases have a specific epidemiological profile, which makes the pre-mortem information of the corpse survey essential, in order to draw reliable conclusions.

According to environmental factors we can state that in our series the most prevalent were co-sleeping and prone position, which represent a high percentage of cases, and where there is a higher co-sleeping incidence at 2 months, and prone position at 4 months.

All of this coincides with the 2 and 4 months of life, when the highest incidence occurs, and endorses the importance of continuing prevention campaigns that, despite having experienced a significant decline after the recommendations issued by the American Academy of Pediatrics in the USA in 1992, it remains steady in recent years<sup>6,7</sup>.

Despite the many investigations carried out on



**Figure 4.** Deceased according to the four seasons of the year.

the pathogenesis of SIDS for decades, none of the proposed pathogenic hypotheses provide conclusive explanations for all cases. The "triple risk theory" is currently the most accepted and states that a vulnerable infant, at a critical stage of development, is subject to a series of external stressor factors (environmental, infectious, among others) to which most infants survive and which do not cause death as such, but may hinder survival in a vulnerable infant.

In recent years, quality improvements in examination of circumstances of death/place of death and formation of multidisciplinary teams to address infant SD have demonstrated that one of the most important risk factors for this type of death is unsafe sleep environment. So it is important to know the percentage of infant SD associated with this circumstance<sup>8,9</sup>.

In all cases a slight predominance of males was observed and there was a markedly less difference in the SIDS cases, (30 males and 23 females). Lack of a clear predominance in the first or precise seasonal

rhythm in our series, as well as the strong association with unsafe sleep environment or increased causes of death explained by the multidisciplinary study led by the Risk Assessment Unit for Sudden Family Death, prove the need to go on working in this field and increase the sample size bringing together different working teams from the various autonomous communities of Spain. This way risk factors would be confirmed and actions will be effectively taken before them.

## CONCLUSIONS

It can be stated that sudden infant death is multifactorial and complex; therefore, requires collaboration of the different medical specialties involved (forensic doctors, pathologists, pediatricians and cardiologists) to be assessed. This way, a multidisciplinary study of sudden infant death provides key information to: set up categories according to the San Diego classification criteria, confirm risk factors to plan prevention measures and strategies, direct the family cardiology study (explain cases of channelopathies and perform diagnosis-early treatment in affected family members) and refer the parents to the risk pregnancy unit.

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