

National Variations in Recent Trends of Sudden Unexpected Infant Death Rate in Western Europe

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Objective To study recent epidemiologic trends of sudden unexpected death in infancy (SUDI) in Western Europe. **Study design** Annual national statistics of death causes for 14 Western European countries from 2005 to 2015 were analyzed. SUDI cases were defined as infants younger than 1 year with the underlying cause of death classified as "sudden infant death syndrome," "unknown/unattended/unspecified cause," or "accidental threats to breathing." Poisson regression models were used to study temporal trends of SUDI rates and source of variation. **Results** From 2005 to 2015, SUDI accounted for 15 617 deaths, for an SUDI rate of 34.9 per 100 000 live births. SUDI was the second most common cause of death after the neonatal period (22.2%) except in Belgium, Finland, France, and the UK, where it ranked first. The overall SUDI rate significantly decreased from 40.2 to 29.9 per 100 000, with a significant rate reduction experienced for 6 countries, no significant evolution for 7 countries, and a significant increase for Denmark. The sudden infant death syndrome/SUDI ratio was 56.7%, with a significant decrease from 64.9% to 49.7% during the study period, and ranged from 6.1% in Portugal to 97.8% in Ireland. We observed between-country variations in SUDI and sudden infant death syndrome sex ratios.

Conclusions In studied countries, SUDI decreased during the study period but remained a major cause of infant deaths, with marked between-country variations in rates, trends, and components. Standardization is needed to allow for comparing data to improve the implementation of risk-reduction strategies. (*J Pediatr 2020;226:179-85*).

udden unexpected death in infancy (SUDI) is defined as the death in an infant less than 1 year old that occurs suddenly and unexpectedly and whose cause is not immediately obvious before investigation. After a complete investigation, including a review of clinical history, an observation of the scene investigation, and an autopsy, SUDI can be attributed to various causes. Death is classified as sudden infant death syndrome (SIDS) when the thorough investigation fails to identify a cause. SIDS must be considered a specific subcategory of SUDI. After the discovery of the prone sleeping position as a major risk factor, massive "back-to-sleep" prevention campaigns were conducted in the early 1990s and led to a huge decrease in SIDS incidence in many countries. However, SIDS and SUDI remain an important cause of infant mortality in countries with high-income economies. 9,11

According to the last international comparisons, SUDI and SIDS rates in Europe varied strongly around mean rates of 39 and 25 cases per 100 000 live births. ^{7,9,12} Austria, Germany, France, and Ireland have particularly high SIDS rates. ⁷ Such high rates led SUDI to be the most common cause of death for infants aged 28 days to 1 year of age. ¹² Previous studies have shown that

male sex is a SIDS risk factor in some countries, but no between-country comparisons have been made on SUDI and SIDS sex ratios. ^{13,14} Besides geographical and sex variations in SIDS rates, some studies have identified important variations in the definition of SUDI, SUDI case investigation protocols, and death coding practices leading to highly variable SIDS/SUDI ratios and impairing reliable international comparisons. ^{9,10,12,15-18}

Despite the public health burden of SUDI, no European analysis of SUDI trends has been performed since 2010. However, such analyses are necessary to justify, inform, and implement future SUDI risk-reduction campaigns in Western Europe countries, notably those with high or increasing SUDI rates. Such comparisons could also help to identify the need to standardize investigation protocols, definitions, and coding practices to improve the reliability of data

ICD-10 International Classification of Diseases, 10th Revision

SIDS Sudden infant death syndrome

SUDI Sudden unexpected death in infancy

WHO World Health Organization

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from routine surveillance practices and allow the evaluation of risk-reduction campaigns. In this context, we aimed to study the geographical, sex, and time variations of SUDI and SUDI subcategories from 2005 to 2015 in Western European countries.

Methods

General Design and Study Data

We performed a secondary analysis of national data on causes of death derived from death certificates. These data were available from the official website of the World Health Organization (WHO) database on mortality for the 14 European countries (Austria, Belgium, Denmark, Finland, France, Germany, Ireland, Italy, Norway, Portugal, Spain, Sweden, the Netherlands, the UK; **Figure 1**) that participated in the Statistical office of the European Union Task Force dedicated to improving the quality and comparability of European cause-of-death statistics. ^{19,20} Available data were

aggregated by year and totally anonymous, so no ethical and regulatory approvals were required. For each country, WHO publishes annual numbers of deaths by causes, based on the *International Classification of Diseases, 10th Revision* (ICD-10) transmitted by national death registries. During the study period, except for Portugal where specific codes were used in 2005, the 14 countries used the ICD-10 for coding the causes of death. WHO provides correspondence tables between the specific Portuguese codes and ICD-10 codes. We used annual numbers of live births from the European Union's statistical office database and transmitted by the national institutes of statistics.²¹ To calculate ratios, rates, and trends in SUDI and SUDI subcategories, we used data from 2005 to 2015, the last year for which data were completely available for all countries.

All deaths in infants younger than 364 days old from 2005 to 2015 were included. By definition, there is no ICD-10 code for SUDI, just as there is no well-established list of ICD-10 codes to define it. We selected a set of ICD-10 codes

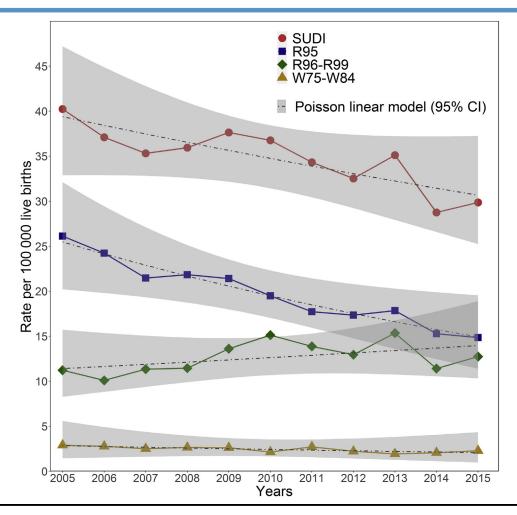


Figure 1. Temporal trends of SUDI and SUDI subcategory rates in 14 European countries (Austria, Belgium, Denmark, Finland, France, Germany, Ireland, Italy, the Netherlands, Norway, Portugal, Spain, Sweden, and the UK) between 2005 and 2015. SUDI was defined as deaths in infants <365 days old with death codes R95 (SIDS), R96-99 (unknown/unattended/unspecified cause), or W75-W84 (accidental threats to breathing) in the ICD-10.

180 de Visme et al

corresponding to death causes found in most cases after SUDI investigations and those previously used for international comparison. 12,22 Thus, we defined SUDI with the ICD-10 cause of death codes R95 "sudden infant death syndrome," R96 "other sudden death, cause unknown," R98 "Unattended death," R99 "other ill-defined and unspecified causes of mortality," W75 "accidental suffocation and strangulation in bed," W76 "other accidental hanging and strangulation," W78 "inhalation of gastric contents," W79 "inhalation and ingestion of food causing obstruction of respiratory tract," W80 "inhalation and ingestion of other objects causing obstruction of respiratory tract," W81 "confined to or trapped in a low-oxygen environment," W83 "other specified threats to breathing," and W84 "unspecified threat to breathing." Because some of these codes accounted for limited numbers, we grouped them in 3 categories: SIDS (R95), unknown/unattended/unspecified cause (R96-R99), and accidental threats to breathing (W75-W84).

To assess and compare the SUDI ratio of infant mortality and those of other death causes, we selected the 2 chapters of the ICD-10 classification with the highest ratios: P00-P96, chapter XVI "Certain conditions originating in the perinatal period" and Q00-Q99, chapter XVII "Congenital malformations, deformations and chromosomal abnormalities." Ratios of SUDI and main causes of death among infant mortality was described overall (0-364 days) and by age subgroups (neonatal death from 0 to 27 days and post-neonatal infant death from 28 to 364 days). SUDI rates were calculated overall and then by SUDI subcategories, sex, age classes.

Statistical Analyses

SUDI rates were calculated as number of deaths for 100 000 live births. SUDI subcategory ratios were calculated by dividing SUDI subcategory cases by overall SUDI (R95, R96-R99 and W75-W84) cases. Poisson regression models of annual death rates were used to assess temporal trends from 2005 to 2015. Annual change in coefficient was calculated with a 95% CI. The statistical significance threshold was fixed at P < .05 and a Rainbow test was used to verify the fit of the models.²³ SUDI sex rate ratios were estimated by median-unbiased estimation (mid-p) and Wald unconditional maximum likelihood estimation. To visualize SUDI rate variations by geographical region, specific rates for 2005-2015 were calculated, and a map of these rates, divided into quartiles, was plotted for 14 European countries. R/RStudio was used for all analyses (R Foundation for Statistical Computing, Vienna, Austria; https://www.R-project.org/).

Results

From 2005 to 2015, in the 14 European countries studied, SUDI accounted for 9.7% of infant mortality, and was the third cause of death after those classified as "certain conditions originating in the perinatal period" (ICD-10 P00-P96) and "congenital malformations or chromosomal abnormalities" (ICD-10 Q00-Q99) which accounted for

52.3% and 25.0% of deaths, respectively (**Table I**). SUDI accounted for 22.2% of infant deaths between 28 and 364 days of age, the second leading cause of death after "congenital malformations or chromosomal abnormalities." In 4 countries (Belgium, Finland, France, and the UK), SUDI was the first cause of death after the neonatal period and for 4 countries (Italy, the Netherlands, Portugal, and Spain), it was the third or fourth cause (**Table I**). Deaths coded P00-P96, Q00-Q99, and SUDI occurred during the neonatal period (0-27 days) in 89.2%, 67.4%, and 30.3% of cases, respectively. For SUDI, deaths occurring during the neonatal period accounted for 88.9% and 91.3% of cases in Denmark and Portugal, respectively, but ranged from 12.8% (Austria) to 46.5% (Sweden) for the 12 other countries.

From 2005 to 2015, 15 617 infants died of SUDI in the 14 Western European countries, for an SUDI rate of 34.9 per 100 000 live births and with a significant decrease (P < .001) from 40.2 to 29.9 (**Table II** and **Figure 1**). The 4 countries in the upper quartile of SUDI rate from 2005 to 2015 (>41.9 per 100 000 live births) were, by decreasing rate, Denmark, France, Belgium, and Germany (**Figure 2**). From 2005 to 2015, 6 countries (Austria, France, Germany, Portugal, Spain, and the UK) showed a significant decrease in SUDI rate, with no significant trend in 7 (Belgium, Finland, Ireland, Italy, the Netherlands, Norway, Sweden), and a significant increase in Denmark (**Table II**).

During 2005-2015, the SIDS/SUDI ratio was 56.7% in the 14 Western European countries, with a significant decrease (P < .001) from 64.9% to 49.7% during this period (**Table II**). The SIDS/SUDI ratio ranged from 6.1% in Portugal to 97.8% in Ireland; 5 countries (Denmark, Italy, the Netherlands, Portugal, and Sweden) had a ratio of less than 50%. We observed a significant decrease in SIDS/SUDI ratio for 5 countries (Belgium, Denmark, France, Germany, and the Netherlands) (**Figure 3**; available at www.jpeds.com), a significant increase for Portugal, and no significant variations for the 8 other countries (**Table II**).

For the 14 Western European countries studied, the rates of SUDI overall, SUDI classified as SIDS, SUDI classified as "unknown/unattended/unspecified cause," and SUDI classified as "accidental threats to breathing" were 39.9, 22.2, 14.4, and 2.7 per 100 000 live births for male infants and 29.5, 15.4, 11.7, and 2.0 for female infants, respectively (**Table III**; available at www.jpeds.com). The male/female sex ratios were 1.4, 1.4, 1.2, and 1.3 for SUDI overall, SIDS, "unknown/unattended/unspecified" cause and "accidental threats to breathing," respectively, with all these sex ratios significantly greater than 1 (P < .001). The sex ratios of SUDI classified as SIDS, SUDI classified as "unknown/unattended/unspecified cause," and SUDI classified as "accidental threats to breathing" were not significantly greater than 1 in 4, 10, and 11 countries, respectively (**Table III**).

Among the 3 SUDI subcategories in 14 European countries, we observed significant (P < .001) negative and positive

2015			% <u>\$</u>	87.2	83.7	1.1	84.5	63.2	72.6	70.9	65.3	75.6	73.1	8.7	81.9	53.5	81.6	2.69		
in 14 European countries 2005-	/s	SUDI	Rank	2	-	7	-	-	7	2	4	က	2	က	က	2	-	2		
			‡%	22.7	28.1	19.9	30.1	29.0	26.8	25.1	8.0	13.6	23.1	12.0	14.7	23.2	23.8	22.2		
		Q00-Q99 [†]	% <u>§</u>	32.1	30.6	19.2	28.6	31.1	36.5	21.0	38.0	27.4	29.4	8.3	45.8	37.6	28.0	32.6		
	28-364 days		Rank	-	7	-	7	7	-	-	-	-	-	-	-	-	7	-		
	28		‡%	32.4	21.6	34.5	29.5	20.6	29.9	39.0	32.2	30.9	27.3	35.9	32.5	28.9	21.9	56.6		
ıfants		٠.	% <u>§</u>	10.9	10.3	3.6	8.0	11.4	12.4	9.3	9.1	9.0	12.7	1.4	12.2	11.1	11.2	10.8		
and ii		*964-004	Rank	3	က	က	က	က	က	က	2	2	က	2	2	က	က	က		
onates		_	‡%	19.3	13.9	15.3	1.1	17.9	19.0	17.2	17.8	19.2	22.5	15.5	19.9	13.7	19.3	18.4		
stribution of the 3 major leading causes of death in neonates and infants in 14 European countries 2005-2015	0-27 days	P00-P96* Q00-Q99 [†]	§%	12.8	16.3	88.9	15.5	36.8	27.4	29.1	34.7	24.4	56.9	91.3	18.1	46.5	18.4	30.3		
			SUDI	SNDI	Rank	3	က	7	က	က	က	က	က	က	က	က	က	က	က	က
			‡%	1.4	2.9	20.6	2.5	8.0	4.9	3.1	1.7	1.5	3.6	7.1	1.6	10.6	2.4	4.2		
			§%	62.9	69.4	80.8	71.4	68.9	63.5	79.0	62.0	72.6	9.07	91.7	57.2	62.4	72.0	67.4		
			Rank	2	7	က	7	7	7	7	7	7	7	7	7	7	7	2		
e 3 ma			‡%	28.5	26.4	18.8	33.5	21.6	25.2	44.0	21.5	27.9	27.7	22.3	21.9	25.2	25.4	24.2		
[able I. Ratios and rank of infant mortality, and age distribution of th			% <u>§</u>	89.1	89.7	96.4	92.0	88.6	97.6	90.7	6.06	91.0	87.3	98.6	87.8	88.9	88.8	89.2		
			Rank	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-		
			±%	0.99	65.3	53.6	57.8	65.7	65.2	50.1	72.8	66.4	65.7	29.7	72.3	57.7	68.9	67.1		
		.P96* Q00-Q99 [†] SUDI	Rank	က	က	က	က	က	က	က	က	က	က	က	က	က	က	က		
			‡%	7.6	11.8	20.5	11.1	14.7	12.1	8.2	3.6	4.6	9.4	7.3	0.9	15.0	9.0	9.7		
	0-364 days		Rank	2	7	7	7	7	7	-	2	2	2	7	7	7	7	2		
	0-364		±%	29.6	24.7	20.6	32.2	21.3	26.8	45.8	24.6	28.7	27.6	23.1	25.4	26.5	24.3	25.0		
rankc			Rank	-	-	-	-	-	-	2	-	-	-	-	-	-	-	-		
os and		P00-P96	‡%	52.3	47.3	49.2	43.2	50.3	50.1	42.5	56.9	54.4	52.9	57.4	54.7	42.5	53.5	52.3		
Table I. Ration			Countries	Austria	Belgium	Denmark	Finland	France	Germany	reland	Italy	The Netherlands	Norway	Portugal [¶]	Spain	Sweden	¥	Total		

*P00-P96, Certain conditions originating in the perinatal period.
1000-Q99, Congenital malformations, deformations, and chromosomal abnormalities.
1Percent among infrant mortality.
SPercent among 0-364 days by death cause category.
¶Data were not available for Ireland from 2005 to 2006 and for Portugal in 2005, 2006, 2009,

2012, and 2013.

trends for SUDI classified as SIDS (R95), "unknown/unattended/unspecified cause" (R96-R99), and "accidental threats to breathing" (W75-W84) (Figure 1). SIDS and "accidental threats to breathing" rates decreased significantly during the study period, but the "unknown/ unattended/unspecified cause" rate increased significantly. The SIDS rate was 19.8 per 100 000 live births, contributing up to 56.7% of SUDI mortality (Table IV; available at www.jpeds.com). "Unknown/unattended/ unspecified cause" and "accidental threats to breathing" rates were 12.7 and 2.4 per 100 000 live births and contributed to 36.3% and 7.0% of SUDI mortality, respectively. SIDS rates significantly decreased in 7 countries (Austria, Belgium, France, Germany, Norway, Spain, and the UK). "Unknown/unattended/unspecified cause" rates significantly increased in 6 countries (Belgium, Denmark, France, the Netherlands, Norway, and Portugal) and significantly decreased in the UK. We observed a significant decrease of "accidental threats to breathing" only in Germany and Italy (Table IV). The contribution of each SUDI subcategory to the overall SUDI rate showed important between-country variations, from 6.1% (Portugal) to 97.8% (Ireland) for SIDS, 2.2% (Ireland) to 83.7% (Denmark and Portugal) for "unknown/unattended/ unspecified cause," and 0% (Ireland) to 19.4% (the Netherlands) for "accidental threats to breathing" (**Figure 4**; available at www.jpeds.com).

Discussion

From 2005 to 2015, SUDI accounted 9.7% of infant mortality in Western European countries, with 15 617 infant deaths during the study period. We found a 25.6% decrease in SUDI rates, from 40.2 to 29.9 per 100 000 live births from the beginning to the end of the study period. This decrease confirms the previous trend observed in Europe from the end of the 1990s.^{7,9,12,24} However, this satisfactory trend on the scale of Western Europe masks more complex national patterns. SUDI national rates varied greatly, with 6-fold differences between countries with the highest and lowest rates, respectively. Moreover, during the study period, trends in SUDI rates showed no reduction in Belgium, Finland, Ireland, Italy, the Netherlands, Norway, and Sweden and a significant increase in Denmark. These marked betweencountry differences may result from variations in different potential contributors: definition of SUDI, SUDI case investigation protocols, and death certifying and coding practices. An important part of the between-country variations in the contribution of each SUDI category to overall SUDI may reflect heterogeneity in how information is collected and analyzed to achieve classification. 17,25 It may also result from disparities in the geographical distributions of modifiable and nonmodifiable SUDI risk factors, such as ethnic origin, socioeconomic status, longitude/latitude data, or genetic data.²⁶⁻²⁸ A last explanation may be found in national variations regarding the implementation level of risk-

182 de Visme et al

S/SUDI ratios in 14 European countries for 2005, 2015, and 2005-2015 trends	2005-2015	SUDI SUDI	Rate* Exponential (β) [95% CI] [†] P value % Exponential (β) [95% CI] [†] P value	0.93 [0.89-0.97] <.001 90.2 0.99 [0.95-1.03]	0.99 [0.96-1.01]	1.04 [1.02-1.07] .002 13.2 0.87 [0.80-0.96]	0.97 [0.92-1.02] 78.2 0.99 [0.94-1.05]	50.6 0.99 [0.98-1.00] .028 52.4 0.96 [0.95-0.97] <.001	0.94 [0.93-0.95] <.001 63.4 0.96 [0.95-0.98]	0.97 [0.92-1.03] . 296 97.8 1.00 [0.94-1.06]	0.99 [0.97-1.01] .365 32.9 1.00 [0.96-1.04]	1.01 [0.98-1.05] .479 43.3 0.94 [0.90-0.99]	0.97 [0.92-1.02] .210 83.2 0.96 [0.91-1.01]	0.90 [0.85-0.95] <.001 6.1 1.25 [1.04-1.51]	0.97 [0.95-0.99] <.001 67.2 0.99 [0.97-1.02]	1.00 [0.96-1.04]	0.97 [0.96-0.98] <.001 61.1 1.00 [0.98-1.01]	34.9 0.98 [0.97-0.98] <.001 56.7 0.97 [0.97-0.98] <.001		
spı		SIDS/SUDI ra	SIDS/SUDI ra	Exponential (eta) [9		_	0.87		_	_	•					_	_	1.00	_	۰.
15 trer	2		%	90.2	67.7	13.2	78.2	52.4	63.4	97.8	32.9	43.3	83.2	6.1	67.2	46.9	61.1	56.7	0 7 0	
005-20	2005-201		P value	<.001	.378	.002	.203	.028	<.001	.296	365	.479	.210	<.001	<.001	.970	<.001	<.001		
for 2005, 2015, and 2		IONS	Exponential (eta) [95% CI] †	0.93 [0.89-0.97]	0.99 [0.96-1.01]	1.04 [1.02-1.07]	_			$\overline{}$		_	_		_		$\overline{}$			
untries			Rate*	27.1	43.1	76.4	56.9	9.09	42.5	28.2	12.0	18.0	25.5	23.2	19.4	37.4	40.1	34.9	7 0 0	
pean co			u	235	292	514	174	4380	3206	182	717	326	167	196	266	456	3442	15 617	•	
I ratios in 14 Euro	2015		SIDS/SUDI Ratio (%)	81.0	32.3	19.1	2.99	53.6	47.0	87.5	28.6	36.8	2.99	21.4	2.69	47.2	54.3	49.7	10 101	
	2005		Rate*	24.9	53.2	80.7	16.2	36.8	36.6	24.4	11.5	1.1	20.4	16.4	15.8	46.1	34.1	29.9	1	
Table II. SUDI rates per 100 000 live births and SID			u	21	92	47	6	280	270	16	26	19	12	14	99	23	265	1193	•	
			SIDS/SUDI Ratio (%)	93.8	83.3	64.7	90.5	64.2	69.3	n/a	27.4	45.2	100.0	n/a	77.2	2.69	26.7	64.9	0 0 0 7 7 0 0	
			Rate*	40.9	45.1	26.4	36.4	49.7	62.7	n/a	13.2	22.4	35.2	n/a	24.5	32.6	46.4	40.2	1000	
DI rate			u	32	24	17	71	382	430	n/a	73	45	20	n/a	114	33	332	1556		
Table II. SU			Countries	Austria	Belgium	Denmark	Finland	France	Germany	Ireland [‡]	Italy	The Netherlands	Norway	Portugal [‡]	Spain	Sweden	¥	Total		

n/a, not applicable.
*Per 100 000 live births.
†Per 100 000 live births.
†Coefficient of a Poisson regression model of the annual change in rate by calendar year.
†Coefficient of a Poisson regression model of the annual change in rate by calendar year.
‡For Ireland, data were not available in 2005 and 2006 and for Portugal in 2005, 2006, 2009, 2012, and 2013; trends were calculated over a 9-year period only for Ireland and a 6-year period for Portugal.
‡For Ireland, data were not available in 2005 and 2006 and for Portugal in 2005, 2006, 2009, 2012, and 2013; trends were calculated over a 9-year period only for Ireland and a 6-year period for Portugal.

reduction campaigns. ^{9,29} Indeed, little is known about current sleeping practices for European children. The few available data suggest that there is not a good implementation level, with, for example, only 72.4% of infants having safe sleeping positions in the Netherlands in 2017, a country with one of the lowest SUDI rates in Western Europe. ³⁰ Thus, thousands of infants are dying each year in Western Europe with the lack of implementation of simple risk-reduction campaigns.

Between-country variations were not limited to SUDI rates and were similarly observed for SIDS/SUDI ratios. These variations were significant (from 6.1% in Portugal to 97.8% in Ireland) and raise the question of major between-country heterogeneity owing to the absence of an international consensus on SUDI definition, post mortem exploration protocols, and/or certifying and coding practices for SIDS. Such variations have been described between regions in the US and France, for post mortem exploration protocols and certifying practices, and between countries for coding practices. 9,12,31,3 Persistent geographical heterogeneity is highly prejudicial because it precludes effective public health efforts using international comparisons and benchmarking between countries with low and high SUDI rates to better implement risk-reduction strategies. The importance of multiagency review is highlighted by several authors and contributes to better-quality investigation and classification. 16-18,33,34 Efforts were made to standardize definitions in other fields of perinatal and pediatric health to allow for international comparisons and guide corrective actions to improve patient outcomes. For example, the Euro-Peristat Project, which has compared perinatal health statistics since 2000 in European countries, highlighted difficulties in comparing data among countries. This project had made proposals for a better use of existing statistics and provided a stimulus to countries to improve the quality of national statistics.^{35,36}

As expected, we observed a significant male/female sex ratio, from 1.2 to 1.4, for SUDI overall, SIDS, "unknown/unattended/unspecified cause," and "accidental threats to breathing." Male infants are well-known to be at greater risk of SIDS. 13,14,24,37 To explain this sex ratio, several hypothesis were proposed: recessive X-linked genetic disorders inducing fatal acute anoxic events or sex differences in the brain, notably brain stem maturation. 38-41 However, we found important between-country variability in this sex ratio, with 4 and 5 countries not sharing it for SIDS and SUDI, respectively. The between-country heterogeneity in sex ratio is complex to analyze. It is not likely related to variations in definitions of SUDI and/or certifying and coding practices. Because male sex is also a risk factor for accidental suffocation and strangulation in bed or shaken baby syndrome, a sex-related difference must be hypothesized in the implementation of risk-reduction strategies and/or in post mortem exploration protocols. 13,14,24,37,42 For these exploration protocols, sex differences may occur in investigation strategies of death causes depending on health professionals or the request of parents. If the several European countries with no significant sex ratio have effectively tackled this

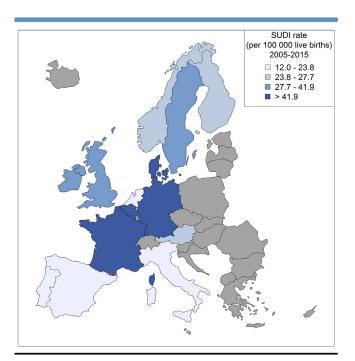


Figure 2. SUDI rates per 100 000 live births for the 2005-2015 period in 14 European countries participating in the Eurostat Task Force.

sex-related excess risk in the implementation of risk-reduction strategies, then thousands of theoretically avoidable male infant deaths have occurred in the other European countries.

Among studied European countries, the distribution of SUDI before and after 28 days of life varied widely. Again, these variations may reflect the differences in certifying and coding practices with no international agreed-upon definition, but another source of variation can be hypothesized: national policies on screening for congenital anomalies and legal gestational age limit for late terminations of pregnancy in some countries such as France. These policies could result in a lower number of "Congenital malformations, deformations and chromosomal abnormalities" after the neonatal period. Finally, the decrease in SIDS/SUDI ratio observed in several countries is generally attributed to a change in the certification and coding of death causes at the country level. 9,12,22,44

Belgium, Finland, France, and the UK were notable regarding SUDI during the study period. These were the only countries where SUDI was the first cause of death after 28 days of life. In the US and probably in other countries, death certificates are classified with a temporary cause of death until the death investigation is completed and the definitive cause of death is attributed. If the death certificate is not appropriately modified after investigation, the temporary cause of death will remain indefinitely, probably most often "unknown/unattended/unspecified cause" or SIDS. One study has shown that 85% of the French SUDI referral centers do not update death certificates with a definitive cause of death. Thus, results of post mortem investigations

and the autopsy do not participate substantially in the official death coding. These inconsistencies create difficulties in the US and France and probably explain why in the 4 countries SUDI was the first cause of death after 28 days of life or more generally why "accidental threats to breathing" causes are rare. 9,45 If death certificates were properly updated, "Congenital malformations, deformations and chromosomal abnormalities" may had a ratio of infant mortality higher than SUDI in these 4 countries. Because we did not directly use death certificates but rather aggregated national statistics on the causes of death (number of deaths by causes), we do not know how many death certificates had more than 1 code documented and how each team dealt with this situation. Finally, the epidemiologic units in this study were countries. A recent study in the US assumed geographic variations in SUDI beyond national borders such as ethnic origin, socioeconomic status, and longitude/latitude data.²⁶

A careful analysis of data from WHO suggests pathways to improve the public health management of SUDI. SUDI remains a major cause of death in Western European infants, but no reliable data are available because of a lack of standardization of the SUDI definition, investigation of SUDI cases, and death certifying and coding practices. Our results suggest the need for a systematic and comparative appraisal of the definitions used in Europe and national registries are needed to gather reliable data on SUDI cases; thus, consensus efforts at the European level are mandatory. Such consensus may be a long-term objective, but it was achieved in other fields such as perinatal health.³⁵ It could allow for standardization and then European comparison of data and benchmarking to improve the implementation and impact of risk-reduction campaigns.

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184 de Visme et al

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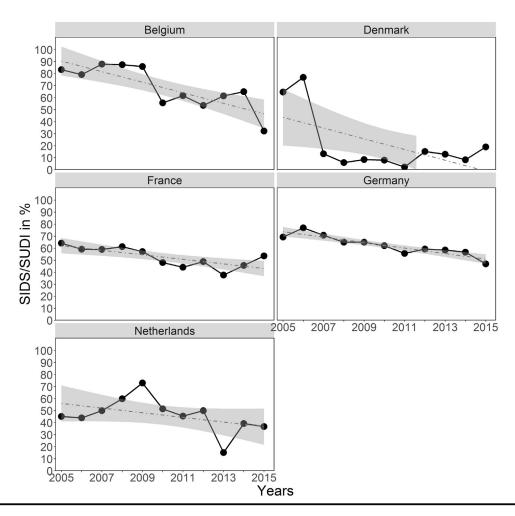


Figure 3. Temporal trends of SIDS/SUDI ratios and Poisson linear model (95% CI) for 5 European countries (Belgium, Denmark, France, Germany, and the Netherlands) between 2005 and 2015.

185.e1 de Visme et al

November 2020

3.5 4.2 2.8

1.1 0.4 1.9

2.4 2.3 2.4

1.5 1.8 1.3

0.8 0.8 0.8

2.8 3.1 2.5

<.001 2.3 2.7 2.0

.604

.440

.215

<.001

<.001

.402

1.5 [0.9-2.6]

0.2 [0.0-1.6]

1.0 [0.4-2.3]

1.4 [0.8-2.3]

0.9 [0.2-3.8]

1.2 [0.9-1.6]

1.3 [1.2-1.5]

.124

.089

.911

.211

.934

.143

<.001

Table III. SUDI and subcategory rates per 100 000 live births by sex and sex ratios in 14 European countries 2005-2015 W75-W84[‡] R96-R991 Rate per 100 Rate per 100 000 Rate per 100 000 Rate per 100 000 000 live live births live births live births births Sex ratio[§] [95% CI] P value M Sex ratio[§] [95% CI] P value Τ M F Sex ratio[§] [95% CI] P value T M F Sex ratio[§] [95% CI] P value Countries T 1.8 2.2 1.4 Austria 27.1 31.4 22.5 1.4 [1.1-1.8] .012 22.1 25.9 18.0 1.4 [1.0-2.0] .024 8.0 0.3 1.4 0.2 [0.0-1.6] .088 1.5 [0.5-4.6] .469 43.2 51.2 34.8 1.5 [1.2-1.7] 26.4 32.1 20.5 1.6 [1.2-2.0] 9.2 10.5 7.9 1.3 [0.9-1.9] 5.5 6.0 5.0 1.2 [0.7-2.0] Belgium <.001 <.001 .158 .490 9.2 11.8 0.8 [0.4-1.4] 1.1 [0.9-1.3] 2.2 3.2 1.1 76.4 81.8 70.8 1.2 [1.0-1.4] 78.2 81.1 75.2 2.9 [0.8-10.5] Denmark .103 8.7 .409 .434 .100 Finland 26.9 30.3 23.4 1.3 [1.0-1.7] 20.0 22.5 17.4 1.3 [0.9-1.9] 3.4 3.3 3.5 1.0 [0.4-2.4] .922 2.3 2.2 2.3 1.0 [0.3-3.0] .936 .094 .188 France 50.6 58.6 42.2 1.4 [1.3-1.5] <.001 25.4 30.3 20.1 1.5 [1.4-1.7] <.001 22.5 24.8 20.0 1.2 [1.1-1.4] <.001 2.6 3.1 2.0 1.6 [1.2-2.1] .003 Germany 42.5 49.0 35.7 1.4 [1.3-1.5] 23.9 28.0 19.6 1.4 [1.3-1.6] <.001 13.2 14.5 11.8 1.2 [1.1-1.4] .004 2.4 2.8 2.0 1.4 [1.0-2.0] .038 <.001 1.2 [0.9-1.6] 0.0 0.0 0.0 Ireland 28.2 30.9 25.5 1.2 [0.9-1.6] .196 27.6 30.0 25.1 .244 0.6 0.9 0.3 2.9 [0.3-27.4] .342 Italy 12.0 13.1 10.9 1.2 [1.0-1.4] .017 4.0 4.5 3.4 1.3 [1.0-1.8] .048 6.4 6.8 6.0 1.1 [0.9-1.4] .277 1.7 1.7 1.6 1.1 [0.7-1.7] .766

.009

.242

.038

.003

<.001

<.001

<.001

6.7 7.0 6.4

3.7 4.3 3.0

4.9 6.3 3.5

19.4 21.2

19.4 18.2

12.3 14.1

13.1 14.4 11.7

17.5

20.6

10.3

1.1 [0.8-1.6]

1.4 [0.6-3.5]

1.2 [0.9-1.7]

1.8 [1.3-2.4]

0.9 [0.7-1.2]

1.4 [1.2-1.6]

1.2 [1.2-1.3]

1.6 [1.1-2.3]

2.1 [1.4-3.2]

2.0 [0.6-6.7]

1.2 [1.0-1.4]

1.6 [1.2-2.2]

1.4 [1.3-1.6]

1.4 [1.4-1.5]

7.7

2.1 4.2 2.1

19.8 26.7

12.2 13.3

.007

.002

.173

.069

<.001

<.001

<.001

9.5

16.5 20.2 12.6

24.1 28.3 19.6

18.8 22.2 15.4

5.9

12.6

11.1

The Netherlands 18.0

Norway

Portugal

Sweden

Spain

UK

Total

†R96-R99, unknown/unattended/unspecified death.

37.4

40.1

±W75-W84, accidental threats to breathing.

§Sex ratio, CI, and P value were computed as rate ratio Wald.

20.5

25.5 31.5 19.1

23.2 25.4 20.9

19.4 22.4 16.3

34.9 39.9 29.5

40.5 34.1

46.1 33.7

15.3

1.3 [1.1-1.7]

1.6 [1.2-2.3]

1.2 [0.9-1.6]

1.4 [1.2-1.6]

1.2 [1.0-1.4]

1.4 [1.3-1.5]

1.4 [1.3-1.4]

F. female: M. male: T. total.

		R95*			R96-R99 [†]	W75-W84 [‡]						
Countries	n (%)	Rate [§]	Exponential (β) [95% CI]	P value	n (%)	Rate [§]	Exponential (β) [95% CI]	P value	n (%)	Rate [§]	Exponential (β) [95% CI] ¶	<i>P</i> value
Austria	212 (90.2)	24.4	0.92 [0.88-0.96]	<.001	7 (3)	0.8	1.17 [0.91-1.50]	.223	16 (6.8)	1.8	0.97 [0.83-1.13]	.689
Belgium	403 (67.7)	29.2	0.92 [0.89-0.95]	<.001	110 (18.5)	8.0	1.34 [1.24-1.45]	<.001	82 (13.8)	5.9	0.98 [0.92-1.05]	.651
Denmark	68 (13.2)	10.1	0.95 [0.88-1.03]	.201	430 (83.7)	63.9	1.06 [1.03-1.09]	<.001	16 (3.1)	2.4	1.02 [0.88-1.19]	.781
Finland	136 (78.2)	21.0	0.96 [0.91-1.02]	.191	23 (13.2)	3.6	1.03 [0.90-1.17]	.704	15 (8.6)	2.3	0.93 [0.79-1.10]	.387
France	2293 (52.4)	26.5	0.95 [0.94-0.97]	<.001	1857 (42.4)	21.5	1.04 [1.02-1.05]	<.001	230 (5.3)	2.7	0.98 [0.94-1.02]	.269
Germany	2033 (63.4)	27.0	0.91 [0.90-0.92]	<.001	979 (30.5)	13.0	1.02 [1.00-1.04]	.062	194 (6.1)	2.6	0.93 [0.89-0.97]	.002
Ireland	178 (97.8)	27.6	0.97 [0.91-1.02]	.247	4 (2.2)	0.6	1.14 [0.77-1.69]	.510	0 (0.0)	0.0		-
Italy	236 (32.9)	4.0	0.99 [0.95-1.03]	.612	383 (53.4)	6.4	1.00 [0.97-1.04]	.856	98 (13.7)	1.6	0.94 [0.88-1.00]	.044
The Netherlands	154 (43.3)	7.8	0.95 [0.91-1.00]	.066	133 (37.4)	6.7	1.08 [1.02-1.14]	.005	69 (19.4)	3.5	1.02 [0.94-1.10]	.668
Norway	139 (83.2)	21.2	0.93 [0.88-0.98]	.006	21 (12.6)	3.2	1.32 [1.11-1.55]	.001	7 (4.2)	1.1	1.01 [0.80-1.28]	.915
Portugal	12 (6.1)	1.4	1.10 [0.91-1.34]	.321	164 (83.7)	19.4	0.86 [0.80-0.91]	<.001	20 (10.2)	2.4	1.13 [0.95-1.34]	.161
Spain	670 (67.2)	13.1	0.96 [0.93-0.98]	<.001	249 (25.0)	4.9	0.98 [0.94-1.02]	.417	78 (7.8)	1.5	0.97 [0.91-1.05]	.451
Sweden	214 (46.9)	17.5	1.00 [0.96-1.04]	.917	233 (51.1)	19.1	1.00 [0.96-1.04]	.920	9 (2.0)	0.7	1.08 [0.88-1.34]	.458
UK	2104 (61.1)	24.5	0.97 [0.96-0.98]	<.001	1082 (31.4)	12.6	0.97 [0.95-0.99]	.005	256 (7.4)	3.0	0.97 [0.94-1.01]	.165
Total	8852 (56.7)	19.8	0.95 [0.94-0.95]	<.001	5675 (36.3)	12.7	1.02 [1.01-1.03]	<.001	1090 (7.0)	2.4	0.97 [0.95-0.99]	.001
Range	6.1-97.8	1.4-29.2			2.2-83.7	0.6-63.9			0.0-19.4	0.0-5.9		

^{*}R95, SIDS.

h95, SIDS.

1R96-R99, unknown/unattended/unspecified cause.

‡W75-W84, accidental threats to breathing.

§Per 100 000 live births.

¶Coefficient of a Poisson regression model of the annual rate by calendar year.

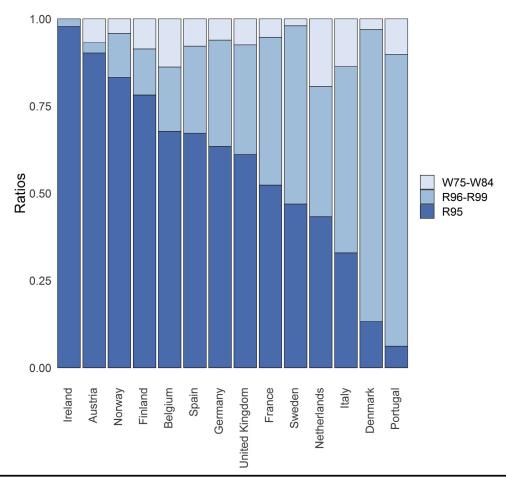


Figure 4. Ratios of SUDI subcategories to total SUDI in the 14 European countries, 2005-2015. SUDI was defined as deaths in infants <365 days old with codes R95 (SIDS), R96-99 (unknown/unattended/unspecified cause), or W75-W84 (accidental threats to breathing) for the cause of death in the ICD-10.