

# Retrospective study of sudden unexpected death of infants in the Garden Route and Central Karoo districts of South Africa: Causes of death and epidemiological factors

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**Background.** Sudden unexpected death in infants (SUDI) is a major contributor to under-5 mortality rates. In attempts to better understand SUDI, an abundance of risk factors has previously been described. However, there is a lack of research pertaining to SUDI and risk factors in South Africa (SA), particularly in rural settings.

**Objective.** To describe the profile of SUDI in rural areas of the Western Cape, SA.

**Methods.** A retrospective analysis was conducted on SUDI cases admitted to the seven mortuaries in the Garden Route and Central Karoo districts (Western Cape) between 1 January 2012 and 31 December 2016.

**Results.** SUDI contributed to 38.56% of all infant deaths and the rate of SUDI was 7.95/1 000 live recorded births. Of the total 5 323 case load, 401 (7.53%) were admitted as SUDI cases. In accordance with other studies, more infant deaths occurred during winter (30.7%) than other seasons and almost all infants demised while sleeping (97.7%). Contrary to other studies, there was a slight female preponderance (54.6%). Symptoms (often mild) of illness prior to demise were reported in 70.2% of infants, but only one-third of these infants' parents/caregivers sought medical attention. Following postmortem investigation, the majority of deaths were due to explained natural causes (93.7%), of which respiratory tract infection was the leading cause of death (74.1% of SUDI admissions). The most prevalent risk factors were: bed-sharing (especially with a smoker), side sleeping, prematurity, exposure to cigarette smoke, maternal alcohol use, unsatisfactory infant weight gain and socioeconomic indicators of deprivation.

**Conclusions.** Overall, the risk factors observed in the rural setting were highly prevalent and were similar to those described in urban areas (both in SA and internationally). Many of these are modifiable and ample opportunity for risk factor intervention was identified, as well as future research opportunities. Most importantly, parents should be educated to not underestimate seemingly mild symptoms in their infants.

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The South African (SA) infant mortality rate (IMR) has steadily declined over the past 5 years and is currently 29/1 000 live births.<sup>[1]</sup> Globally, the IMR ranges from an average of 3/1 000 live births in the European Union to an average of 53 /1 000 live births in sub-Saharan Africa.

In SA, infant deaths contribute to about 75% of under-5 deaths.<sup>[2]</sup> The five leading causes of infant mortality are reported by Statistics South Africa as: 'respiratory and cardiovascular disorders specific to the perinatal period, influenza and pneumonia, disorders related to length of gestation and fetal growth, intestinal infectious diseases and other disorders originating in the perinatal period'.<sup>[3]</sup> Unnatural causes account for only about 3.4% of infant deaths.

Sudden unexpected death in infancy (SUDI) refers to the sudden and unexpected death of a child under 1 year of age, for which there is no immediate apparent cause. Thus, SUDI is a collective term, which refers to the circumstances under which the infant has died, and it is not a diagnosis. In SA, these cases are, by law, admitted to medicolegal mortuaries for postmortem examination.<sup>[4,5]</sup>

The pathologist will eventually classify the death as: (i) natural: deaths due to medically explained natural pathology; (ii) unnatural: accidental deaths (for instance asphyxia due to overlay), or infanticide

(for instance in homicidal smothering); (iii) sudden infant death syndrome (SIDS): deaths which remain unexplained after exhaustive investigation;<sup>[6]</sup> and (iv) undetermined: deaths where no adequate cause of death was found, but in which a shortfall in death scene investigation or ancillary investigations prohibits the pathologist from making a confident diagnosis of SIDS.

Epidemiological studies on SUDI have identified numerous intrinsic and extrinsic risk factors for these deaths. Furthermore, commonalities between risk factors for true SIDS cases and medically explained SUDI have been described.<sup>[7-9]</sup> Epidemiological studies were mainly conducted in developed countries in Europe (including the UK), Australia, New Zealand, USA and, to a lesser extent, in Asia. Studies from the African continent, and from rural areas, are lacking.

## South Africa and SUDI research

In 1989, Molteno *et al.*<sup>[10]</sup> described early childhood deaths in Cape Town and found the rate of 'cot deaths' in Cape Town to be 3.8/1 000 live births. This was followed by a long pause in published research, until Du Toit-Prinsloo *et al.*<sup>[11]</sup> published their findings on a retrospective descriptive study (2011). This

was conducted on SUDI cases admitted between 2000 and 2004 to the Pretoria and Tygerberg medicolegal mortuaries. Another multicentre, retrospective study followed, conducted at the Tygerberg, Bloemfontein, Durban, Johannesburg and Pretoria medicolegal mortuaries, on cases from 2005 to 2009.<sup>[12]</sup> Neither of these studies reported a SUDI rate (SUDI/1 000 live births). These two studies revealed the variance in the extent of postmortem examinations and ancillary investigations performed, both between and within institutions. The need for the implementation of a standardised death scene investigation and comprehensive autopsy protocol was emphasised. Dempers *et al.*<sup>[13]</sup> found the establishment of such a protocol attainable during a feasibility study at Tygerberg medicolegal mortuary. To the best of our knowledge, such a protocol has not yet been implemented at provincial or national level.

## Objective

The above local studies have all been conducted at large academic mortuaries, serving metropolitan communities. The lack of SUDI data in the rural populations of SA was the motivation for undertaking this study. The aim of this study was to describe the epidemiology of SUDI in the Garden Route (formerly known as 'Eden') and Central Karoo districts of the Western Cape (WC) Province, SA, and therefore to identify the burden as well as the 'profile' of the infant most at risk for SUDI in these regions. Furthermore, we aimed to establish how the characteristics of SUDI in the study area compared with those of urban regions in our country and other countries. The hypothesis was that risk factors for SUDI and SIDS, as previously described in the literature, are highly prevalent in both the true SIDS and the medically explained SUDI cases in these areas. The objective was to conduct a descriptive study on SUDI cases admitted to medicolegal mortuaries in this geographical area.

## Methods

### Study setting

The municipal districts of the Garden Route and of the Central Karoo are two separate, but geographically adjacent, entities in the WC Province.<sup>[14-16]</sup> Together, these districts formed the setting for this research study. Although these districts occupy almost half (48%) of the province's surface area, they are less densely populated – only about 11% of the population of the WC Province resides in the study area. The population density of the Garden Route District is 25 inhabitants/km<sup>2</sup>. Many of the inhabitants of the sparsely populated Central Karoo (2 inhabitants/km<sup>2</sup>) belong to farming communities. Thus, the population density of the study area is in stark contrast to the densely populated Cape Town Metropole (1 639 inhabitants/km<sup>2</sup>).

Regarding health parameters, at the time of the study, the neonatal mortality rate of the Central Karoo (10/1 000 live births) was more than double that of the Cape Town Metropole (4.3/1 000 live births) and the prevalence of malnourished children under the age of 5 ( $n=11/100\ 000$ ) almost tenfold that of the Cape Town Metropole ( $n=1.8/100\ 000$ ). One-fifth of babies in the Central Karoo were underweight at birth. The Central Karoo also had a considerably higher rate of teenage pregnancies than Cape Town. The Garden Route District's health parameters were more favourable than those of the Central Karoo, but less favourable than those of the Cape Town Metropole.

### Forensic pathology services within the study setting

The adjoining municipal districts of the Garden Route and Central Karoo form one functional forensic pathology district. Seven of

the 16 Forensic Pathology Service (FPS) laboratories (medicolegal mortuaries) of the WC are located in the Garden Route and Central Karoo districts. Ranging from holding facilities to a referral centre, these mortuaries are located in George, Mossel Bay, Riversdale, Knysna, Oudtshoorn, Laingsburg and Beaufort West.

When an unnatural death is suspected, FPS is called to the death scene, where scene observations are documented, some family history is recorded and the body is collected. These observations are documented on a standard 'FPS006(b) form' which is based on the international guidelines by the Centers for Disease Control and Prevention (CDC).<sup>[17]</sup> At the mortuary, the pathologist will review the available information, where it may become evident that symptoms to explain a natural disease process were indeed present prior to death. In these cases, and if there is no suspicion of foul play involved, it is not mandatory for FPS to investigate further. However, in cases where a natural disease process is not evident from the history and/or foul play is suspected, a postmortem examination is performed. This involves evisceration of the body and dissection and macroscopic examination of the organ blocks. The autopsy may be terminated if an adequate natural cause of death is found (a limited autopsy).

Ancillary investigations are not routinely performed if the cause of death is concluded by dissection and macroscopic examination. Ancillary investigations that are readily available, and practical to conduct in this region, include histology, on-site rapid HIV antibody testing and toxicology. Postmortem radiology, in the form of full-body digital X-ray (LODOX-Statscan., SA) was available for a limited period only.

### Study design

A retrospective approach was followed to describe the profile of SUDI in the Eden and Central Karoo districts of the WC Province. All SUDI admissions to the FPS mortuaries within these areas during a 5-year period between 1 January 2012 and 31 December 2016 were reviewed. The FPS Case Register (FPS R003) of each mortuary was searched manually for all infant (a child under the age of 1 year) admissions. The cases were cross-checked against the electronic case list which was extracted from the FPS Business Information Management System (FPS BIM).

Children >1 year old, proven stillbirths and non-viable fetuses were excluded from the study. Cases which were initially admitted to the mortuaries as SUDI cases, but where a medical practitioner subsequently issued a death registration form, declaring the death to be due to natural causes, were excluded, since these cases did not undergo a postmortem examination/autopsy and no medicolegal report was compiled.

### Data collection

A data collection form for numerous variables was constructed (see Appendix 1: <http://www.sajch.org.za/public/files/1729.pdf>). This included demographic information, circumstances surrounding death, clinical history and autopsy findings and the risk factors for SUDI, which were identified during the literature review.

Data were collected from the original case files in the archives of each mortuary and/or from their electronic equivalents stored on Livelink (Open Text, Canada). The main documents in the files from which the data were collected were the FPS006(b), the FPS001 (Incident Log Form) and FPS002 form (Scene script), the Road to Health booklet and the pathologist's postmortem report. Other documents included the declaration of death and the notice of death/stillbirth forms as well as the SA Police Service (SAPS)

180 form, which is completed by the SAPS Officer at the scene. Data were captured electronically using the EpiData (EpiData; Denmark) tool.

### Data analysis

The captured data were exported from EpiData to Excel (Microsoft Corp., USA) and to the Statistica-64 (StatSoft, USA) software programme. Excel was used for performing basic statistics and to create visual presentation of the data. Statistica-64 was used to perform more advanced statistics. Hypothesis testing for categorical variables was performed by using the  $\chi^2$  test, with a *p*-value of <0.05 considered to be statistically significant. Missing data were excluded from statistical analyses.

For statistical analyses, infant death ages were categorised into: early neonatal deaths (<7 days old), and late neonatal deaths (7 - 28 days old). Postneonatal cases were categorised as follows: 4 - 8 weeks old; 8 - 12 weeks old; 3 - 6 months old; 6 - 9 months old and 9 - 12 months old. For the purpose of this study a day was regarded as starting at 07h00. Thus, deaths between 07h00 and 19h00 were regarded as daytime deaths and deaths between 19h00 and 07h00 as night-time, including early-morning, deaths. Laingsburg mortuary's data were reported individually for basic statistics. However, because of the small case number at this mortuary (*n*=7) its data were merged with those of Beaufort West mortuary for the performance of tests for associations. Thus, for significance testing the two mortuaries in the Central Karoo were combined.

Cases where the cause of death was indicated as SIDS/probable SIDS were discussed with a senior specialist pathologist serving these areas. If such a case did not at least have a death scene investigation, a complete autopsy and histological examination, the cause of death was reclassified, for the purposes of this study, to 'undetermined', as these cases did not fit the minimum criteria for SIDS, which is by definition a diagnosis of exclusion.<sup>[6]</sup> Further, while the cause of death was initially captured exactly as per the postmortem report, cases that were merely reported as 'natural' were further subcategorised if the type of natural death was clear from the information in the file (these were mainly cases where there was a clear history of respiratory tract symptoms and where an internal autopsy was subsequently not performed).

Each district's recorded live births and mortality reports were retrieved from Statistics SA's website to calculate the estimated IMR for each year (number of infant deaths per 1 000 live births).<sup>[18,19]</sup> The recorded live births and SUDI case totals were used to calculate the estimated SUDI rate for each year (number of infant deaths per 1 000 live births).

### Approval and ethical considerations

Approval to access the FPS's case files, both in the form of the original case file and the electronic file, was granted by the Directorate: Forensic Pathology Services, Department of Health, WC. Ethical approval for this study was obtained from the Institutional Review Board (HREC ref. no. 036/2017).

### Results

A total of 5 323 cases (adults and children) were admitted to the medicolegal mortuaries in the Garden Route District (*n*=4 322) and the Central Karoo District (*n*=1 001) of the WC. SUDI cases accounted for 7.53% of total admissions and 91.34% (*n*=401) of all infant admissions. (see Appendix 1: <http://www.sajch.org.za/public/files/1729.pdf> for the number of cases per mortuary

per annum). An overall decline in SUDI numbers from 2012 to 2016 was observed, with an associated overall decline in the annual SUDI rate (see Appendix 1: <http://www.sajch.org.za/public/files/1729.pdf>).

The average IMR in these two districts over the 5-year study period was ~20.62/1 000 recorded live births. The average SUDI rate was ~7.95/1 000 live births. On average, 38.56% of all infant deaths in these districts were SUDI cases.

The majority (54.61%; *n*=219) of infants were female. A significant association between female deaths and autumn was found (*p*=0.001). Winter was the season with the most deaths (30.67%; *n*=123) and also the only season in which more males (*n*=67) than females (*n*=56) demised. The least number of deaths occurred during summer (18.95%; *n*=76) (Fig. 1).

Regarding the time of day, the majority of deaths (62.59%; *n*=251) occurred during the night and early hours of the morning (19h00 - 07h00). Mossel Bay was the only mortuary where significantly more infant deaths occurred during the day than during the night (*p*=0.05).

Fig. 2 illustrates the uneven distribution of SUDI between the days of the week. This variable was not significantly different between the mortuaries (*p*=0.77). Sunday was the day with the most deaths (27.68%) and an association was found between death on Sundays and respiratory tract infections (*p*<0.0001).

Almost half of infants (46.13%; *n*=185) demised during weekends (Fridays 19h00 - Mondays 07h00). This tendency of death during weekends (as opposed to weekdays) was statistically significant in the farming communities (*p*=0.02) and informal settlements (*p*=0.02), but not for infants from formal settlements (Fig. 3).

The vast majority of infants demised before the age of 6 months (89.28%; *n*=358) and more than half of the infants demised before the age of 12 weeks (61.60%; *n*=247) (Fig. 4). There was no significant difference in the distribution of male and female deaths by age category (*p*=0.76).

Most infants (90.77%; *n*=364) died in their own homes while 4.24% (*n*=17) died at a healthcare facility and 3 died en route to a healthcare facility. Two infants died in their cots at childcare facilities. In more than half (58.85%; *n*=236) of all SUDI cases, symptoms of illness were present prior to the death. The symptoms varied in severity, as perceived by parents, and also in time period present before demise (less than a full day to more than 2 weeks). Respiratory tract symptoms were highly prevalent, with rhinorrhoea/blocked nose the most frequently reported symptom (76.27% of symptomatic infants), followed by coughing (67.80%). Other prevalent symptoms were fever (48.31% of symptomatic infants), diarrhoea (32.63%), vomiting (25.42%), restlessness (19.92%) and listlessness (16.95%). Only one-third (33.47%; *n*=79) of symptomatic infants were taken to a healthcare facility (including pharmacies, clinics, hospitals and other) for medical attention.

Fig. 5 shows the distribution of the cases by broad category of death. Medically explained, natural causes of death were found in the vast majority of cases (94.01%; *n*=377). Respiratory tract infections were the leading cause of death, contributing to almost three-quarters of all SUDI cases (74.06%; *n*=297). Structural cardiac abnormalities and inborn errors of metabolism, among others, contributed to deaths related to congenital disorders. Only 6 cases were eventually categorised as SIDS. Satisfactory history, scene observations, a complete autopsy and histology were performed, and revealed no specific cause of death. A further 12 cases were categorised as undetermined, either because no cause of death was found at autopsy, but histology was not performed, or because of the

**Table 1. Frequency of known risk factors for SUDI observed in this study**

	<i>n</i>	% of cases (for which the risk factor was known)	Availability of information (%)
Antenatal and obstetric risk factors ( <i>N</i> =401)			
Maternal smoking	163	55.82	72.82
Low birthweight (<2 500g)	182	54.50	83.29
Prematurity	155	47.55	81.30
Maternal alcohol use/abuse	98	46.23	52.87
Maternal HIV	62	38.50	40.15
No antenatal care (or late care)	65	33.33	48.63
Prolonged hospitalisation after birth	61	30.35	50.12
Advanced maternal age (>35 years)	57	14.54	97.76
Grand multiparity (≥5 live births)	45	14.20	79.05
Young maternal age (<19 years)	47	11.99	97.76
Maternal hypertension/pre-eclampsia	46	11.47	43.89
Maternal drug abuse	7	3.47	50.37
Twin pregnancy	6	1.59	100
Sleep-related risk factors ( <i>n</i> =297)*			
Not sleeping in a cot/baby bed	285	93.90	99.33
Co-sleeping	249	92.56	90.57
Side sleeping	130	71.75	90.57
Co-sleeper smoking	132	53.01	83.84
Prone sleeping	30	9.67	90.57
Sociodemographic risk factors ( <i>N</i> =401)			
Mother unemployed	183	87.98	51.87
Mother did not attend/complete high school	164	86.90	41.90
Single mother	258	86.87	74.06
Deceased infant was 3rd child (or later)	170	53.63	79.05
Informal housing	111	30.33	91.27
Farm	41	11.20	91.27
Postnatal and miscellaneous risk factors ( <i>N</i> =401)			
Exposure to cigarette smoke	185	62.93	73.32
Poor weight gain	66	38.37	42.89
Missed most/all routine clinic visits	20	9.35	53.37
Missed some/all immunisations	40	16.19	61.60
Previous SUDI of a sibling	23	7.26	79.05

\*297 infants demised during a sleeping period.

possible contribution of neglect to the death of the infant. Accidental causes of death, due to unsafe sleeping conditions, were found in a minority of cases (1.75%; *n*=7).

In 78 (26.26%) of the respiratory-related deaths, an element of gastroenteritis/acute diarrhoea was stated as a contributory factor. Thus, gastroenteritis/acute diarrhoea was either the primary cause of death or a contributor to death in almost one-quarter of all SUDI cases (24.44%; *n*=98).

An overall association between age at death and cause of death was observed ( $p<0.0001$ ). Significantly more respiratory-related deaths than other deaths were seen in infants older than 3 months ( $p<0.0001$ ). Deaths due to congenital disorders and unexplained and unnatural deaths were absent in infants older than 6 months (Fig. 5). The majority of deaths attributed to congenital disorders occurred in infants younger than 8 weeks (70.59%; *n*=12).

A full internal autopsy was performed in more than half of cases (52.62%; *n*=211). In 8.48% (*n*=34) of all SUDI cases ancillary investigations were performed. Limited autopsies, terminated once an adequate cause of death was found, were rarely performed (1.25%; *n*=5).

An external examination only was performed in 46.88% (*n*=188) of cases. Histology was the most frequently performed ancillary investigation and was performed in 33 cases. Rapid HIV antibody screening tests were performed on site in 3 cases. Postmortem radiology, in the form of full-body digital X-ray (LODOX-Statscan, SA) was used in two cases, confirming the suspicion of pneumonia and deeming an internal autopsy unnecessary. Genetic evaluation was requested in only 1 case. In 2 cases the postmortem report was missing and no indication about the extent of postmortem investigations was available.

### Risk factors

The case studies also involved the description of the prevalence of well-known risk factors for SUDI (Table 1). As indicated in the table, comprehensive information on all cases was sometimes lacking. Maternal smoking was the most prevalent antenatal risk factor, with more than half of infants exposed to maternal smoking *in utero*. Antenatal smoking exposure was not equally distributed between the areas ( $p=0.004$ ), with significantly more infants exposed in the



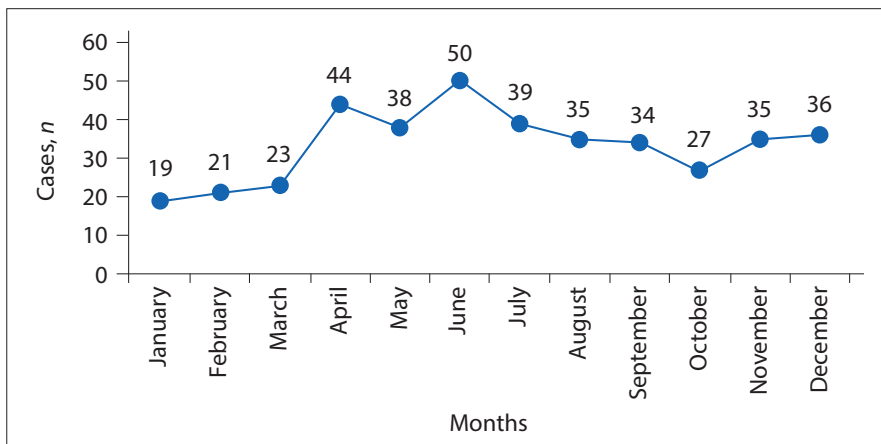


Fig. 1. Distribution of SUDI by month. January: 4.74%; February: 5.24%; March: 5.74%; April: 10.97%; May: 9.48%; June: 12.47%; July: 9.73%; August: 8.73%; September: 8.48%; October: 6.73%; November: 8.73%; December: 8.98%.

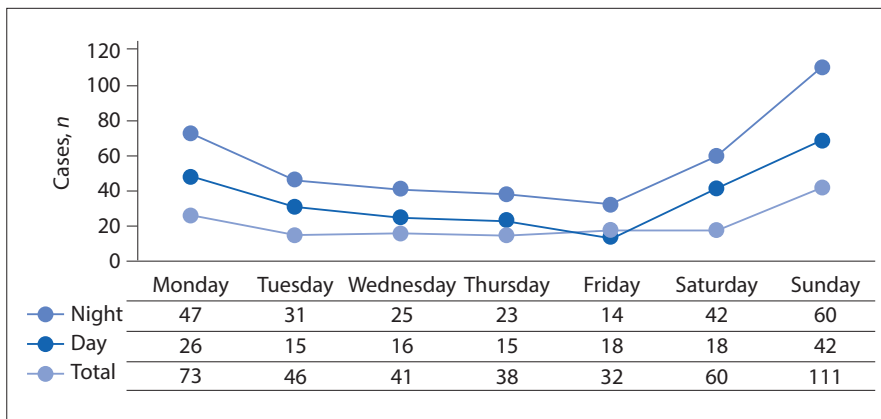


Fig. 2. Distribution of cases by day of the week and time of day. Almost half of the infants demised at weekends, with Sundays being the most common day of the week.

Central Karoo ( $p=0.0002$ ) and Riversdale ( $p=0.03$ ) areas. Low birthweight and prematurity were also highly prevalent.

While information on maternal alcohol use in pregnancy was only available in 52.87% of cases, the data available showed 46.23% of mothers regularly consumed alcohol during pregnancy. An association between maternal alcohol use and area was found ( $p=0.0001$ ), with a tendency for alcohol-exposed cases to come from the Central Karoo ( $p=0.0002$ ) and Riversdale ( $p=0.03$ ) areas once again.

Only 10 infants (3.37%) were sleeping in their own cot when they demised. More than 90% were co-sleeping when they demised, most of them sharing a bed with one adult (49.44% of all cases where the information was available) or two adults (21.19%). More than half of the co-sleeping adults were smokers. Most infants were placed in a side sleeping position and a few in a prone position. The most common position in which the demised infants were found was on their side (48.15%), followed by a supine position (38.15%) and the minority were

found prone (11.11%). In 7 cases the position of the infant was described as wedged or overlain. However, not all 7 of these deaths were eventually found to be asphyxia related.

Undesirable socioeconomic conditions, such as maternal unemployment, incomplete school education and single motherhood were highly prevalent. On the other hand, most infants resided in formal homes in residential areas, with about one-third living in informal housing structures and about a tenth on farms. More than half of infants were the third or later child born to the mother.

The most prevalent risk factor in the postnatal period was regular exposure to cigarette smoke after birth. There was a significant association between passive smoking and respiratory-related deaths and also deaths due to gastroenteritis/acute diarrhoea. Generally, adherence to routine clinic visits was noted, and most infants' immunisations were up to date. Information about previous deaths in the family was available in almost 80% of cases and revealed

that in 7.26% of cases, the same mother had experienced a previous case of SUDI.

## Discussion

Statistics on SUDI and SIDS are lacking in developing countries, including SA.

In countries where SUDI and SIDS rates are available, SIDS rates have declined since the 'back to sleep' campaigns of the 1980 - 1990s.<sup>[20]</sup> The rate of true SIDS (~0.63 deaths/1 000 live births) in our study was remarkably lower than that described for Cape Town in 1989.<sup>[10]</sup> However, our study's SIDS rate was higher than recent international SIDS rates, which vary from 0.05/1 000 live births in Sweden to 0.39/1 000 live births in the USA (2012 - 2014).<sup>[20]</sup>

In most countries with available data, SUDI rates have also followed a downward trend and currently range between 0.14/1 000 live births (the Netherlands, 2008 - 2010) and 0.88 (USA, 2014). In stark contrast is our study's high SUDI rate of ~7.95/1 000 live births. That said, international comparison of rates for SIDS and SUDI, as well as the recognition of trends over time, may be thwarted by diagnostic preference, differences in diagnostic coding and diagnostic shift owing to the evolution of the definitions for SIDS and SUDI over time.<sup>[20]</sup>

There is no ICD-10 code for SUDI, as it is not a diagnostic entity. These cases are masked by their final diagnosis (e.g. pneumonia) within SA's mortality data. Hence the medicolegal mortuary data of the Garden Route and Central Karoo proved crucial in revealing that 38.56% of all infant deaths in these areas occurred suddenly and unexpectedly, with no immediate obvious cause of death.

SUDI cases formed 7.53% of the total case burden of this district's medicolegal mortuaries. The inconsistency of the referral of SUDI cases to medicolegal mortuaries in different areas of SA poses a challenge to the comparison of case numbers.<sup>[11,21]</sup> Nevertheless, the proportion of SUDI observed in this study was higher than the average SUDI case load for the five large medicolegal centres serving metropolitan SA communities.<sup>[12]</sup> In addition, several important aspects were discovered pertaining to these SUDI cases, and these results are discussed in comparison with three key studies: (i) an SA multicentre study, containing the findings of five large academic mortuaries,<sup>[12]</sup> (ii) a German study (GeSID)<sup>[6]</sup> and (iii) a British study (CESDI).<sup>[8]</sup>

In clear disparity with the European studies, in this study 93.77% of deaths were medically explained. This proportion was

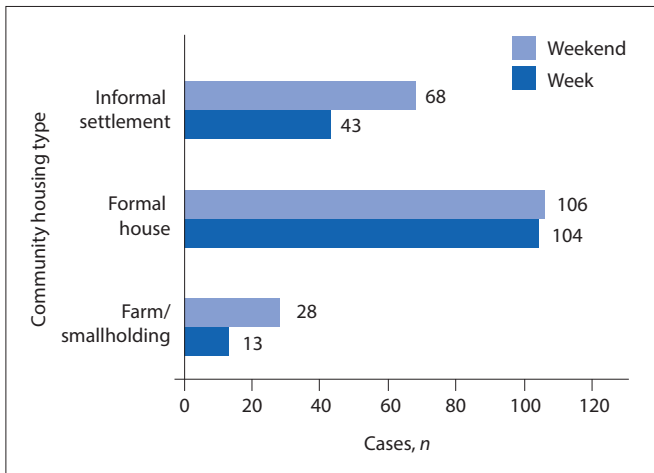


Fig. 3. Distribution of cases by community housing type and time period of the week. Significantly more deaths were observed on weekends as opposed to weekdays for informal settlements and farming communities.

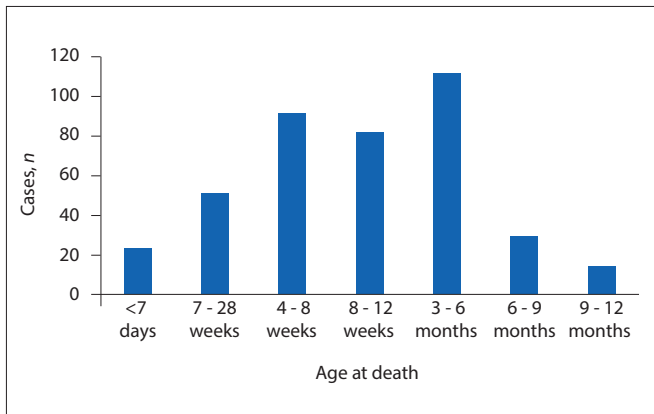


Fig. 4. Distribution of cases by age category at death. Early neonatal deaths: 5.74%; late neonatal deaths: 12.72%; 4 - 8 weeks: 22.69%; 8 - 12 weeks: 20.45%; 3 - 6 months: 27.68%; 6 - 9 months: 7.23%; 9 - 12 months: 3.49%.

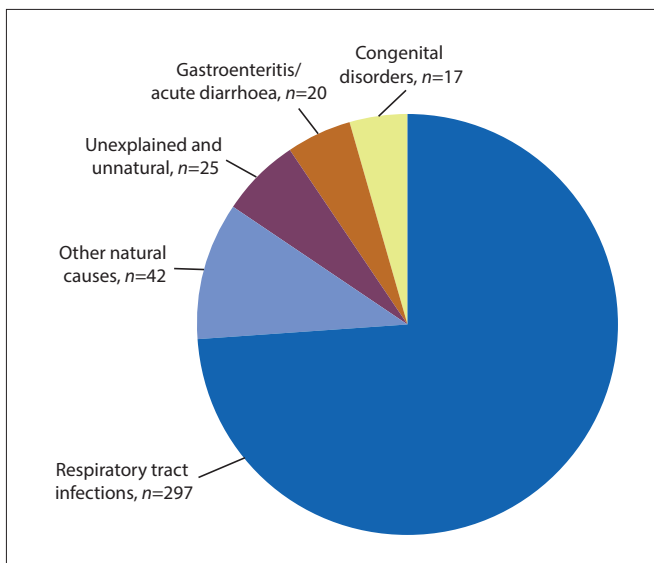


Fig. 5. Distribution of cases by cause of death. Respiratory tract infections: 74.06%; other natural causes of death: 10.47%; unexplained (including SIDS) and unnatural causes: 6.23%; gastroenteritis/acute diarrhoea: 4.99%; congenital disorders: 4.24%.

also higher than the SA multicentre study's average (75.5%). Only 1.50% of SUDI admissions were attributed to true SIDS, much lower than the SA multicentre study (8.7%). However, the range of SIDS-categorised cases varied profoundly between these five mortuaries (0.0 - 14.9%), which might also indicate differences in diagnostic criteria, or variance in opinion between pathologists and centres. Undetermined deaths amounted to 3.0% of the SUDI admissions in this study, while the SA metropolitan mortuaries figures ranged between 1.7% and 37.2%. The European studies classified more than 80% of their cases as SIDS and left no cases undetermined.

Different explanations can be offered for these statistical differences. Although SA is regarded as an upper middle-income country, its extreme level of inequity<sup>[22]</sup> and relatively high IMR<sup>[1]</sup> stand alone compared with the other country studies of SUDI. Additionally, the functioning of the healthcare system in SA is further challenged by the implications of historical injustices, as well as by social and political factors. These include, among others, high rates of violence, and racial and gender discrimination, as well as lack of leadership and human resources.<sup>[23,24]</sup> When comparisons with countries with different socioeconomic profiles are made, it is therefore possible that some differences in findings are merely reflections of this socioeconomic gap. Almost a third (30.3%) of infants lived in informal homes, and 11.2% lived on farms.

Additional true SIDS cases could have been concealed in this study, in both the medically explained and the undetermined groups. The true rate of SIDS in this area will not be known if ancillary investigations, at least histology and virology, are not routinely performed. To this end, there has been a call for the development of a standardised protocol for the investigation of all SUDIs in SA.<sup>[12]</sup> Such a protocol would be comparable with international standards, and would thus promote the comparison of data within SA and also internationally.<sup>[13,25]</sup> This would include a detailed, standardised death scene investigation of high quality, by trained forensic pathology officers (including detailed collection of information on the infant and including concepts such as doll re-enactment).<sup>[11,26]</sup> This would be followed by complete autopsy with histology, virology, bacteriology and possibly molecular/genetic investigations as the minimum ancillary investigations. However, for the purposes of FPS in SA, it is not mandatory to establish if SUDI cases were truly SIDS or a medically explained death, as both are regarded as natural deaths.<sup>[4]</sup> Furthermore, the cost implication of additional investigations on all SUDI cases, as well as the additional burden on pathologists serving this large geographical area, would be considerable.

Similar to the literature, most deaths in this study occurred during the night and early hours of the morning. Mossel Bay was the only mortuary to have more deaths during the day than the night. Perhaps this finding is a result of the relatively small sample size from Mossel Bay or it may be a feature of death recording in the area.

A disproportionate number of infants died during weekends: 29% more than expected if there was an even distribution, with a prominent peak occurring on Sundays. Fridays were the only days where the total deaths during the daytime slightly outnumbered that of the night/early morning. This weekend predominance was statistically significant in the farming communities and the informal settlements. A few other studies have also described this weekend phenomenon.<sup>[27-30]</sup> The current thoughts to explain this are changes in the infant's routine over weekends, in social activities of the caretaker and in medical attention-seeking behaviour or diminished access to healthcare services over weekends.<sup>[28,29,31]</sup> Prospective, case-control studies might shed some light on these

findings.

There was an extremely high rate of co-sleeping (92.6%) and 46.6% of these shared the bed with more than one other person, of whom at least one was an adult. However, an earlier study on the sleeping habits of infants in the Cape Peninsula revealed a high rate of co-sleeping (94%) in the general infant population.<sup>[32]</sup> This suggests that additional factors may be more significant than mere co-sleeping itself. Many studies have found that the risk of co-sleeping and SIDS/SUDI was only significant if the co-sleepers were smokers.<sup>[33-36]</sup> In this study 53.01% of co-sleepers smoked. Others studies found that the association between co-sleeping and SIDS/SUDI was particularly high in infants younger than 4 months,<sup>[34,37]</sup> when the co-sleeper had used drugs or taken alcohol before going to bed,<sup>[38]</sup> and also over weekend periods.<sup>[39]</sup> Indeed, smoking exposure was highly prevalent in this study, where 55.8% of infants' mothers smoked during pregnancy. This percentage was substantially higher than that of the SIDS and medically explained group of both European studies. However, it should be noted that a shortcoming of this study was the lack of quantification of the mother's smoking, where the other studies considered smoking exposure only when the mother smoked at least 10 cigarettes per day.

There was a significant association between passive smoking and respiratory-related deaths and also deaths due to gastroenteritis/acute diarrhoea. While passive smoking is only one aspect of indoor air pollution, the contribution of indoor air pollution might be further explored. Limited research has described a possible association between sudden infant death and air pollution.<sup>[40-42]</sup> In SA, the burning of fuels (solid/liquid) inside homes is common<sup>[31-34]</sup> and the contribution of this to sudden infant death may offer a research opportunity.

In all the studies used for comparison, respiratory-related deaths (such as bronchopneumonia and pneumonitis) were the leading cause of medically explained deaths; however, this study's rate was far higher than the others (74.06% of all SUDI cases and 79.00% of all the medically explained SUDI cases). Most infants in this study had some symptoms of illness prior to death. Of the symptomatic infants, the most prevalent symptoms were those of a mild respiratory tract infection, which was consistent with the comparison studies. Many of the infants had had such symptoms for a time period of 7 - 14 days.

Only a third of infants were taken for medical attention, which is also in keeping with the literature, which explains that parents often do not recognise the seriousness of an illness in their infant.<sup>[8]</sup> Parents should be educated on the significance of minor respiratory tract symptoms in these high-risk infants, and medical attention-seeking behaviour should be enhanced. Risk scoring tools for parents, by which they can be alerted to the severity of symptoms of a systemic illness, are needed.<sup>[8]</sup> The inclusion of such a tool in the Road to Health booklet of SA infants can potentially save infant lives.

The majority of infants in this study (61.1%) were either premature and/or low-birthweight infants. Many of these infants spent a prolonged time in hospital after birth. This might provide an opportunity for clinicians to educate the parents of these vulnerable infants on SUDI risk factors and the reduction thereof. Further, as poor infant weight gain has been identified as a risk factor for SUDI, great attention should be given to growth, especially of the low-birthweight infants. The importance of frequent health checks should be emphasised and meticulous assessing of weight gain should be performed. This may require a critical review of growth assessment tools for preterm infants.<sup>[47]</sup> Overall, many risk factors for SIDS/SUDI were highly prevalent in the study cohort.

### Study limitations

As with most retrospective studies, this study was limited by the information available in the case files. While not unique to our study area, the missing information once again highlights the lack of standardised death scene investigation and documentation thereof, which was recently pointed out at a large academic medicolegal mortuary in Cape Town.<sup>[48]</sup>

The descriptive, retrospective nature of this study precludes the calculation of the prevalence of the SIDS/SUDI risk factors in the entire infant population and poses a limitation to the interpretation of these findings. Prospective, case-controlled studies, with matched controls, would be more informative and would provide for more accurate statistical inference. However, studies in this region, which covers such a vast area, could be a challenge in terms of cost and time constraints.

### Conclusions

SUDI is the main contributor of infant mortality in many countries.<sup>[20]</sup> While this entity is well studied in many developed countries, there is a shortfall of knowledge in less-developed countries. This study was the first to be situated in a rural, non-academic setting in SA, and contributed new data to the burden of SUDI and associated risk factors in these areas. In the Garden Route and Central Karoo, 80 infants die suddenly and unexpectedly each year and these deaths contribute to almost 40% of all infant deaths.

The vital registration system in SA does not capture SUDI deaths *per se*, which is a disadvantage. By including the circumstances of death (for instance 'SUDI' or 'pedestrian vehicle accident') on the death notification form, the data on these cases would become available. Fortunately, with the recent implementation of a Child Death Review project in the Western Cape, these deaths are now further discussed and investigated by multidisciplinary panels in which FPS, paediatricians, Department of Social Development and the National Prosecuting Authority, among others, are represented.<sup>[21]</sup> In this way, the social circumstances of siblings are attended to as needed. The provision of healthcare services in these cases is also evaluated in this process, and refined where indicated. Main outcomes are to prevent future deaths and to ensure that cases of child neglect are identified and dealt with speedily and efficiently.

Two questions often remain: Was this death preventable? Why did a seemingly healthy infant die so suddenly and unexpectedly of a natural cause, with nil or only minor symptoms present?

The profile of the infant at risk for SUDI in the Garden Route and Central Karoo districts is that of a premature baby, exposed to ante- and postnatal smoking, under the age of 6 months, who develops symptoms of a seemingly minor respiratory tract infection. This infant would be routinely sharing a sleeping surface with more than one other person, of whom at least one is a smoker. The infant would be put to bed, by parents unaware of the potential fatal cascade of pathophysiological events occurring in their child. This would probably happen at a weekend, in the winter, and the infant would not wake the next morning. This natural death, being preceded by symptoms, will very likely not receive a complete autopsy, and will be categorised as an explained death, of which a bronchopneumonia would be the most probable diagnosis.

Importantly, many previously described risk factors for SIDS were highly prevalent in this study and many infants were inundated with a co-mixture of risks. This contributes to the theory of a common pool of risk factors and clinical characteristics for SIDS and medically



explained sudden infant death. There is abundant opportunity for risk factor intervention in this area, of which the reduction of antenatal smoking and passive smoking might have the greatest impact. Furthermore, by continuing to strive towards universally implemented protocols and by collaborating internationally on research priorities, we can also make a significant contribution to the scientific community studying these young deaths.<sup>[49]</sup>

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