

Infant Sleep Hazards and the Risk of Sudden Unexpected Death in Infancy

Melanie Estelle MacFarlane, MSc¹, John M. D. Thompson, PhD¹, Jessica Wilson, MSc (Hons)¹, Beverley Lawton, MBChB², Barry Taylor, FRACP³, Dawn E. Elder, FRACP⁴, Nick Baker, FRACP⁵, Gabrielle K. McDonald, MPH³, Jane Zuccollo, FRCPA⁶, Martin Schlaud, MD, PhD⁷, Peter Fleming, MB, PhD⁸, and Edwin A. Mitchell, FRACP¹

Objective To examine the effects of infant sofa-sleeping, recent use by caregivers of alcohol, cannabis, and/or other drugs, and bed type and pillows, on the risk of sudden unexpected death in infancy (SUDI) in New Zealand.

Study design A nationwide prospective case–control study was implemented between March 2012 and February 2015. Data were collected during interviews with parents/caregivers. “Hazards” were defined as infant exposure to 1 or more of sofa-sleeping and recent use by caregivers of alcohol, cannabis, and other drugs. The interaction of hazards with tobacco smoking in pregnancy and bed sharing, including for very young infants, and the difference in risk for Māori and non-Māori infants, also were assessed.

Results The study enrolled 132 cases and 258 controls. SUDI risk increased with infant sofa-sleeping (imputed aOR [IaOR] 24.22, 95% CI 1.65–356.40) and with hazards (IaOR 3.35, 95% CI 1.40–8.01). The SUDI risk from the combination of tobacco smoking in pregnancy and bed sharing (IaOR 29.0, 95% CI 10.10–83.33) increased with the addition of 1 or more hazards (IaOR 148.24, 95% CI 15.72–1398), and infants younger than 3 months appeared to be at greater risk (IaOR 450.61, 95% CI 26.84–7593.14).

Conclusions Tobacco smoking in pregnancy and bed sharing remain the greatest SUDI risks for infants and risk increases further in the presence of sofa-sleeping or recent caregiver use of alcohol and/or cannabis and other drugs. Continued implementation of effective, appropriate programs for smoking cessation, safe sleep, and supplying safe sleep beds is required to reduce New Zealand SUDI rates and SUDI disparity among Māori. (*J Pediatr* 2022; ■:1–9).

Sudden infant death syndrome (SIDS) is a diagnosis of exclusion for infant deaths that remain unexplained despite a thorough investigation comprising a detailed clinical history, review of circumstances, scene examination, and full autopsy.¹ However, deaths once certified as SIDS often are labeled as accidental suffocation and strangulation in bed (*International Classification of Diseases* code W75), or ill-defined and unspecified (R99).² The term SUDI (sudden unexpected death in infancy) is commonly used in 1 of 2 ways.³ The first describes the unexpected death of an infant younger than 1 year of age and usually during sleep. After investigation, some of these deaths are explained, for example, previously unrecognized cardiac anomalies and metabolic abnormalities. The second way combines 3 *International Classification of Diseases* diagnoses: R95 (SIDS), W75, and R99, which together, capture deaths previously labeled as SIDS. We use SUDI in this second way, consistent with its use by the Centers of Disease Control and Prevention, the American Academy of Pediatrics, and the Ministry of Health in New Zealand.

Bed sharing, when an infant is asleep on the same sleep surface as 1 or more other sleeping individuals,⁴ is known to increase the risk of SUDI, especially among very young infants,⁵ and features in more than one-half of all SUDI cases in New Zealand.^{5,6} The combination of tobacco smoking in pregnancy and bed sharing creates a dangerous interaction that increases an infant’s risk of SUDI.⁵

There is contention that bed sharing is only a risk if smoking or some other hazard is present.⁷ However, there is evidence that bed sharing continues to present some risk of SUDI in the absence of smoking and other hazards,⁵ including infant sleeping on a sofa, and recent use by caregivers of alcohol and/or cannabis and other drugs. Sofas have been shown to be unsafe for infant sleep,^{8,9}

From the ¹Paediatrics: Child and Youth Health, University of Auckland, Auckland, New Zealand; ²Centre for Women’s Health Research, Victoria University of Wellington, Wellington, New Zealand; ³Women’s and Children’s Health, University of Otago, Otago, New Zealand; ⁴Department of Paediatrics and Child Health, University of Otago, Wellington, New Zealand; ⁵Department of Paediatrics, Nelson-Marlborough Hospital, Nelson, New Zealand; ⁶National Perinatal Pathology Service (NPPS), Auckland City Hospital, Auckland, New Zealand; ⁷Department of Epidemiology and Health Monitoring, Robert Koch Institute, Berlin, Germany; and ⁸Centre for Academic Child Health, Bristol Medical School, University of Bristol, United Kingdom

Sponsors of this study include the Health Research Council of New Zealand (<https://hrc.govt.nz>), which funded the initial feasibility study, and this case–control study (11/261 to E.M.), and the PhD development award (17/496 to M.M.); and Cure Kids (<https://curekids.org.nz>), which supported E.M. and J.T. The authors declare no conflicts of interest.

Portions of this study were presented the online conference handbook during the New Zealand Paediatric Society 71st Annual Scientific Meeting, November 20–22, 2019, Auckland, New Zealand.

0022-3476/\$ - see front matter. © 2022 Elsevier Inc. All rights reserved.
<https://doi.org/10.1016/j.jpeds.2022.01.044>

DHB	District Health Board
IaOR	Imputed aOR
SIDS	Sudden infant death syndrome
SUDI	Sudden unexpected death in infancy

particularly when shared with 1 or more sleeping individuals, and especially when the individuals have used alcohol or drugs recently.⁷⁻⁹ Infants asleep on a sofa, alone or with others, are potentially at risk of suffocation from overlay or becoming wedged against the sofa or the narrow, sloping seats.^{7,8} Recent use of alcohol and drugs by caregivers also is associated with increased SUDI risk, especially when combined with bed sharing on any surface.⁷ Cannabis is used relatively commonly among pregnant and nonpregnant women, and men and women at conception and postnatally.¹⁰⁻¹²

We investigated the risk of SUDI from hazards related to the infant sleep environment and the sleep environment itself, including for very young infants. The interaction of hazards with tobacco smoking in pregnancy and bed sharing were assessed, as was the difference in risk of hazards for indigenous New Zealand Māori and non-Māori infants.

Methods

The New Zealand case-control SUDI Nationwide Study was conducted between March 2012 and February 2015, inclusive. The study, which has previously been reported in detail,⁵ investigated infant sleep practices, antenatal and postnatal health, and infant living situations in relation to SUDI.

Cases

All cases were SUDI deaths in New Zealand during the study period. All such deaths are investigated by a coroner and include a police investigation, a SUDI liaison (trained investigator) interview, and full autopsy. Cause of death (SIDS, ill-defined and unspecified, and accidental suffocation, and strangulation in bed) was determined by an expert group after all investigations had concluded, independent of the coronial system. Deaths from other causes were excluded.⁵

Controls

Controls were randomly sampled after frequency-matching to cases by obstetric hospital of birth, sex, maternal ethnicity (as recorded in hospital birth registers, which was confirmed during the interview), and age at interview aligned with the distribution of New Zealand SUDI cases between 2003 and 2007. Due to the control group being frequency matched in proportion to infants in the 2003-2007 SUDI cohort in respect of obstetric hospital of birth, sex, maternal ethnicity, and infant age at death, the control group in this study was of greater risk of SUDI compared with a nationwide population-based representative sample.

The control group was selected to reflect the characteristics of the population most at risk from SUDI and allows the identification of more subtle differences between cases and controls. A District Health Board (DHB) midwife or mortality review committee coordinator identified the randomized infants from hospital birth registers according to these criteria.

Data Collection

SUDI liaison personnel collected data during face-to-face interviews with mothers, or other caregivers, responsible for the infant during the last sleep (cases) or nominated sleep (controls). Each control infant was allocated a nominated sleep time to reflect the distribution of the time of the last sleep of cases in previous years. If the infant was asleep at the nominated time, this sleep was explored during the interview. If the infant was awake at the time of the nominated sleep, the sleep explored during the interview was either the sleep period prior to, or after this nominated time. The direction was randomly attributed as part of the selection criteria.

Interviews lasting 90-120 minutes occurred at participants' homes and followed detailed, health-focused questionnaires that differed for cases and controls only in reference to the last or nominated sleep. Objective measures, general observations, and scene reconstruction photographs (not part of this report) also were obtained for the study, plus a separate dataset for each SUDI case's coroner. Mothers self-identified their ethnicity during the interviews.¹³ Where possible, relevant child health, obstetric, and medical records were reviewed.

Recruitment

All families received an information letter inviting them to participate. Separate letters were developed for cases and controls. The SUDI liaison telephoned control families within a fortnight of sending the letter to provide information and arrange an interview, if possible. In one DHB area (of relatively low socioeconomic status), controls were first contacted by telephone to enable timely confirmation of contact details. Case families were telephoned as soon as possible. Case families received their letters at the interview.

Explanatory Variables

The primary variables hypothesized to be associated with potentially hazardous situations within the sleep environment were infant sofa-sleeping for the last or nominated sleep; and recent use of alcohol and/or cannabis and other drugs, within 24 hours of the last (cases) or nominated (controls) sleep by the person who placed the infant to sleep and/or cared for them during sleep.

The number of standard alcoholic drinks consumed in the 24 hours preceding the nominated or last sleep (for controls and cases respectively) was collected. One standard drink in New Zealand contains 10 g of alcohol, for example, 100 mL of table wine (12.5% alcohol), or 330 mL of beer (4% alcohol).¹⁴

We examined these 3 hazards' variables to estimate the risk they pose individually and collectively ("combined hazards") to all cases and controls. We also stratified the sample by infant age to assess the effects of the hazards, tobacco smoking in pregnancy, and/or bed sharing on the risk of SUDI among very young case and control infants (<3 months).

When determining the presence of hazards, an affirmative response ("yes") to any hazard variable was coded as "yes"; a hazard variable was coded as "no" if participants responded "no" to each of the hazards, or they were otherwise coded as

missing. The presence of hazards was also assessed for any interaction with tobacco smoking in pregnancy, bed sharing or both. We define bed sharing as 1 or more individuals asleep with an infant on the same sleep surface, regardless of surface type.

Other sleep environment-related variables collected were type of bed, presence of pillows and their position in relation to the infant, how firmly bedding was tucked in, and whether other items were present when the infant was placed in a cot (crib) or cot-type bed eg, cot bumpers or toys. A cot-type bed is of similar shape to a cot and is specifically designed—or intended—for infant sleep. It includes cots, bassinets, Moses baskets, and safe infant sleep beds (wahakura and Pēpi-pods^{15,16}). Mattress softness was measured by passing a 2-kg weight through the aperture of a specially constructed board and measuring the depression in the mattress.¹⁷ Using the re-enactment dolls, mattress softness measurements in millimeters were taken in the location of the infant's chest when they were placed to sleep, and again where they were found unresponsive (cases) or awake (controls). All weights, boards, and dolls in each research kit were the same weight and size for consistency. Data on tobacco smoking in pregnancy and bed sharing were collected during interviews. Infant ethnicity was based on mothers' self-identified ethnicity at the time of interview.

Sample Size

All infant deaths in New Zealand from March 2012 to February 2015 that met the criteria for SUDI were eligible for the study. Based on previous SUDI mortality data,¹⁸ 210 cases were expected across the 36-month study period and a sample of 420 controls. The number of SUDI cases was less than expected due to a decrease in unexpected infant deaths during the study period.¹⁹

If a risk factor had a prevalence of 20% in the control population, the study could detect an OR of 1.73 or greater with a power of 80% at a level of significance of 5%. However, fewer than expected controls participated in the study. To counter this, if a selected control could not be enrolled, a further control was selected.

Imputation and Sensitivity Modeling

Datasets of 90 participants (cases: $n = 59$, 44.7%; controls: $n = 31$, 12.0%) had missing data related to the hazards, tobacco smoking in pregnancy, and/or bed sharing. We used the multiple imputation procedure in SAS (version 9.4, SAS Institute)²⁰ to create 100 imputed datasets based on the distribution of characteristics and patterns of responses from the available data using infant sex, infant age, last bed sleep, bed sharing, breastfed, maternal smoking in pregnancy, maternal age, infant birth weight, parity, marital status, alcohol use, and drug use. Logistic regression models were run for each of the 100 imputed datasets and the MIANA-LYZE procedure was used to combine variable estimates and produce one set of odds ratios. The MI and MIANA-LYZE procedures assume that data are missing at random.

Data Analyses

Univariable and multivariable analyses examined relationships between the relevant variables and the risk of SUDI. Unconditional multivariable logistic regression was used to adjust for potential confounders and determine the presence of interactions. The association of risk factors with SUDI was estimated using OR with a 95% CI. Analyses were carried out in SAS (version 9.4, SAS Institute). Logistic regression was applied using the SAS logistic procedure.

The covariates from the previous publication⁵ were included in the multivariable models. These were maternal ethnicity, maternal age, parity, marital status, infant age, infant sex, birth weight, breastfeeding status, multiple birth status, sharing the parental bedroom, position placed to sleep, tobacco smoking in pregnancy, and bed sharing. We also examined interactions between the hazards, smoking in pregnancy, and bed sharing and the effects of the hazards on Māori and non-Māori. SUDI mortality rates were calculated using the number of live births between 2012 and 2014 inclusive.²¹ Although the controls were frequency matched based on the distribution of cases from 2003 to 2007, there is no guarantee that this will reflect the distribution of these variables within the cases during the study period. Thus, the variables that are used for frequency matching are controlled for in analyses. The ORs for these variables are not reported as they do not reflect the true level of risk associated with these variables, including them in the model simply helps to adjust for imbalances caused by the change in the distribution of these variables from that expected. These variables are not analyzed in relation to hazards, except for purposes of stratification by ethnicity.

Ethics

The study received ethics approval from the Central Region Ethics Committee (CEN/11/09/045) and locality assessment was received from all DHBs. All parents/caregivers provided informed, written consent.

Results

During the 3-year study, 303 infant deaths were referred to a coroner, of which 137 (45%) were classified as SUDI and eligible for the study. Of these, 96% ($n = 132$) of families were interviewed, as were 40% ($n = 258/649$) of selected control infants/mothers. Cases' interviews occurred between 24 hours and 7 days of death. Of the 391 controls who did not participate, 182 were uncontactable, and 209 refused, or accepted but were not available to interview. Approximately one-half the infants in the case and control groups were Māori (49.2% and 52.3%, respectively). Overall, the national SUDI rate during the study period was 0.76 in 1000 live births.⁵ The national SUDI rates for Māori and non-Māori were 1.41 in 1000 and 0.53 in 1000 live births, respectively.²²

As reported previously, infant exposure to the combination of bed sharing and tobacco smoking in pregnancy statistically significantly increases the risk of SUDI.⁵ This

dangerous combination was reconfirmed in the present study through analyses of imputed and unimputed data (imputed aOR [IaOR] 29.0, 95% CI 10.10-83.33; unimputed aOR 22.67, 95% CI 6.0-85.66). There was no statistically significant increase in risk associated with bed sharing on its own (IaOR 1.79, 95% CI 0.59-5.43; unimputed data aOR 1.96, 95% CI 0.46-8.41). Alone, tobacco smoking in pregnancy was associated with a significantly increased risk of SUDI (IaOR 2.53, 95% CI 1.04- 6.11), which was not evident in the unimputed analysis (aOR 0.84, 95% CI 0.24-2.96).

Risks from Exposure to the Hazards

Sofa Sleeping. Although uncommon (cases: n = 11, 8.7%; controls: n = 1, 0.4%), an infant sleeping on a sofa during the last or nominated sleep was associated with a significantly increased risk of SUDI (IaOR 24.22, 95% CI 1.65-356.40; unimputed aOR 38.43, 95% CI 2.13-692.50) (Table I). All 12 infants who slept on a sofa did so during the night or early morning. Of the 11 infants who died while asleep on a sofa, 8 were sharing the sofa with their mother at the time.

Recent Use of Alcohol by Caregivers. Recent use of alcohol by caregivers during the last sleep was reported in one-quarter of cases for whom data were available (n = 18, 25.4%) compared with 7.2% of controls (n = 17). Caregivers of cases who drank alcohol consumed a median of 4 standard drinks compared with caregivers of controls for whom 1 standard drink was the median. Alcohol consumption by caregivers did not result in a statistically significant increased risk of SUDI in the imputed analysis (aOR 2.40, 95% CI 0.90-6.39), but it did reach statistical significance in the unimputed analysis (aOR 3.04, CI 1.04-8.89) (Table I).

Recent Use of Cannabis and Other Drugs by Caregivers.

Few caregivers reported recent use of cannabis and other drugs (cases: n = 6, 8.0%; controls: n = 1, 0.4%), and usage was not statistically significantly associated with SUDI (IaOR 6.92, 95% CI 0.59-81.02; unimputed aOR 6.41, 95% CI 0.44-94.26) (Table I).

Risk from the Combined Hazards. The exposure of infants to 1 or more of the hazards (sofa-sleeping and recent use by caregivers of alcohol and of cannabis and other drugs) was reported in 43.2% of cases (n = 32) and 8.4% (n = 19) of controls (only 3 cases and no controls were exposed to 2 or more hazards). Exposure to 1 or more of these factors was associated with a significant increase in the risk of SUDI (IaOR 3.35, 95% CI 1.40-8.01; unimputed aOR 5.39, 95% CI 2.16-13.46) (Table I).

Effects of the Hazards on Māori and Non-Māori

Māori infants exposed to 1 or more hazards, specifically sofa-sleeping, or recent use by caregivers of alcohol and/or cannabis and other drugs, were at a statistically significant increased risk of SUDI in the multivariable imputed analysis (aOR 6.51, 95% CI 1.67-25.46), whereas this did not reach statistical significance in non-Māori infants (aOR 1.92, 95% CI 0.54-6.86, respectively). However, a test for an interaction of hazards in Māori participants compared with non-Māori participants did not reach statistical significance (imputed $P = .12$, unimputed $P = .14$) (Table II).

Risk from the Hazards in Combination with Bed Sharing and Tobacco Smoking in Pregnancy

There was no statistically significant increased risk of SUDI associated with the presence of 1 or more of sofa-sleeping

Table I. Effects of the hazards (sofa-sleeping and/or recent use by caregivers of alcohol and/or cannabis and other drugs) among cases and controls

Hazards	Cases (%) N = 132	Controls (%) N = 258	Unimputed		Imputed	
			Univariable OR (95% CI)	Multivariable* aOR (95% CI)	Univariable OR (95% CI)	Multivariable* aOR (95% CI)
Sofa-sleeping during the last sleep (cases) or nominated sleep (controls)	(Missing = 5)	(Missing = 0)				
Yes	11 (8.7)	1 (0.4)	24.37 (3.11-190.99)	38.43 (2.13-692.50)	23.50 (3.00-184.09)	24.22 (1.65-356.40)
No	116 (91.3)	257 (99.6)	Reference	Reference	Reference	Reference
Alcohol in last 24 h	(Missing = 61)	(Missing = 22)				
Yes	18 (25.4)	17 (7.2)	4.38 (2.11-9.06)	3.04 (1.04-8.89)	3.26 (1.59-6.68)	2.40 (0.90-6.39)
No	53 (74.6)	219 (92.8)	Reference	Reference	Reference	Reference
Drugs (cannabis and other drugs†) in last 24 h	(Missing = 57)	(Missing = 197)				
Yes	6 (8.0)	1 (0.4)	21.57 (2.55-182.15)	6.41 (0.44-94.26)	12.48 (1.49-104.76)	6.92 (0.59-81.02)
No	69 (92.0)	248 (99.6)	Reference	Reference	Reference	Reference
Hazards combined (1 or more hazards)	(Missing = 58)	(Missing = 31)				
Yes	32 (43.2)	19 (8.4)	8.34 (4.32-16.09)	5.39 (2.16-13.46)	4.88 (2.58-9.20)	3.35 (1.40-8.01)
No	42 (56.8)	208 (91.6)	Reference	Reference	Reference	Reference

*Covariates used in the multivariable analysis include maternal ethnicity, marital status, parity, baby sex, twin, breastfed, sharing parental bedroom, position placed to sleep, maternal age at birth of infant, baby birth weight, baby's age.

†Cases: methadone n = 2; and herbal highs n = 1; controls: nil other drugs reported.

Table II. Effects of the hazards (sofa-sleeping and/or recent use by caregivers of alcohol and/or cannabis and other drugs) on Māori and non-Māori controls

Māoris	Māori cases (%) n = 62	Māori controls (%) n = 135	Unimputed		Imputed	
			Univariable OR (95% CI)	Multivariable aOR (95% CI)	Univariable OR (95% CI)	Multivariable aOR (95% CI)
Hazards	(Missing = 22)	(Missing = 20)		<i>P</i> = .14		<i>P</i> = .12
Yes	19 (47.5)	7 (6.1)	13.96 (5.22-37.37)	10.02 (2.46-40.88)	8.53 (3.31-22.01)	6.51 (1.67-25.46)
No	21 (52.5)	108 (93.9)	Reference	Reference	Reference	Reference
Non-Māoris	Non-Māori cases (%) N = 70	Non-Māori controls (%) N = 123	Unimputed		Imputed	
			Univariable OR (95% CI)	Multivariable aOR (95% CI)	Univariable OR (95% CI)	Multivariable aOR (95% CI)
Hazards	(Missing = 28)	(Missing = 23)				
Yes	13 (38.2)	12 (10.7)	5.16 (2.07-12.88)	2.76 (0.66-11.54)	3.09 (1.28-7.48)	1.92 (0.54-6.86)
No	21 (61.8)	100 (89.3)	Reference	Reference	Reference	Reference

and/or recent use by caregivers of alcohol and/or cannabis and other drugs (IaOR 1.99, 95% CI 0.43-9.24; unimputed aOR 2.36, 95% CI 0.47-11.85). Similarly, the risk of bed sharing combined with 1 or more hazards did not reach statistical significance (IaOR 1.68, 95% CI 0.11-26.04; unimputed aOR undefined). However, these results are based on very low numbers (Table III).

In the imputed and unimputed analyses for infants exposed to tobacco smoking in pregnancy plus 1 or more hazards (IaOR 13.02, 95% CI 2.85-59.37; unimputed aOR 12.31, 95% CI 2.62-57.83) (Table III), a statistically significant increased SUDI risk was identified.

Although the combination of bed sharing, tobacco smoking in pregnancy, and the hazards increased the SUDI risk dramatically (IaOR 148.24, 95% CI 15.72-1398.31; unimputed aOR 159.89, 95% CI 15.28 to >999.999), this estimate is based on a single control (0.4%) reported to have been exposed to this combination, compared with approximately 20% of cases (Table III).

Risk for Very Young Infants

Infants younger than 3 months of age are at a statistically significant increased risk of SUDI when sleeping in a bed

sharing situation even when not exposed to tobacco smoking in pregnancy and hazards (IaOR 10.65, 95% CI 1.55-73.11; unimputed aOR 15.95, 95% CI 1.33-191.14). The risk escalates further with the addition of tobacco smoking in pregnancy (IaOR 190.57, 95% CI 24.53-1480.30; unimputed aOR 180.77, 95% CI 11.10 to >999.999). Very young infants exposed to bed sharing, tobacco smoking in pregnancy, and 1 or more of sofa-sleeping and recent use by caregivers of alcohol, cannabis, or other drugs, are at extremely high risk of SUDI (IaOR 450.61, 95% CI 26.84-7593.14), although this is based on small numbers (Table IV).

Imputed vs Unimputed Analyses

The estimated ORs using imputed and unimputed data in multivariable analyses were all similar, except one. The OR associated with smoking in pregnancy showed no increased risk in the unimputed analysis but did show a 2-fold increased risk in the imputed analysis.

Bed Type

Infants who slept in an adult bed, or on a sofa or other surface, either alone or with another person, all showed a similar level of increased SUDI risk compared with infants who slept

Table III. The risk of SUDI in combination with bed sharing, tobacco smoking in pregnancy, and hazards (sofa-sleeping and/or recent caregiver use of alcohol and/or cannabis and other drugs)

Hazards	Unimputed data			Imputed data		
	Cases (%) n = 73 (Missing = 59)	Controls (%) n = 227 (Missing = 31)	Multivariable aOR (95% CI)	Cases (%) n = 132 (Missing = 0)	Controls (%) n = 258 (Missing = 0)	Multivariable aOR (95% CI)
Bed sharing only	4 (5.5)	24 (10.6)	1.96 (0.46-8.41)	11 (8.3)	27 (10.5)	1.79 (0.59-5.43)
Smoking only	9 (12.3)	60 (26.4)	0.84 (0.24-2.96)	25 (18.9)	68 (26.4)	2.53 (1.04-6.11)
Smoking + bed sharing	18 (24.7)	13 (5.7)	22.67 (6.0-85.66)	38 (28.8)	16 (6.2)	29.0 (10.10-83.33)
Hazards (1 or more)	3 (4.1)	11 (4.8)	2.36 (0.47-11.85)	4 (3.0)	12 (4.7)	1.99 (0.43-9.24)
Hazards + bed sharing	1 (1.4)	2 (0.9)	<0.001 (<0.001 to >999.999)	2 (1.5)	2 (0.8)	1.68 (0.11-26.04)
Smoking + hazards	8 (11.0)	5 (2.2)	12.31 (2.62-57.83)	10 (7.6)	6 (2.3)	13.02 (2.85-59.37)
Smoking + hazards + bed sharing	20 (27.4)	1 (0.4)	159.89 (15.28->999.999)	24 (18.2)	1 (0.4)	148.24 (15.72-1398.31)
None	10 (13.7)	111 (48.9)	Reference	19 (14.4)	126 (48.8)	Reference

Table IV. Very young infants (<3 months) and the risk of SUDI in combination with bed sharing, tobacco smoking in pregnancy, and hazards (sofa-sleeping and/or recent caregiver use of alcohol and/or cannabis and other drugs)

	Unimputed data			Imputed data		
	Cases (%) available n = 37 Missing = 27	Controls (%) available n = 119 Missing = 20	Multivariable aOR (95% CI)	Cases (%) n = 64 Missing = 0	Controls (%) n = 139 Missing = 0	Multivariable aOR (95% CI)
Bed sharing only	4 (10.8)	9 (7.6)	15.95 (1.33-191.14)	7 (11.1)	10 (7.1)	10.65 (1.55-73.11)
Smoking only	2 (5.4)	33 (27.7)	0.88 (0.06-12.75)	6 (9.2)	38 (27.0)	1.59 (0.23-10.72)
Smoking + bed sharing	10 (27.0)	6 (5.0)	180.77 (11.10 to >999.999)	24 (37.0)	6 (4.3)	190.57 (24.53-1480.30)
Hazards (one or more)	0 (0.0)	3 (2.5)	<0.001 (<0.001 to >999.999)	0 (0.3)	4 (2.8)	NA
Hazards + bed sharing	0 (0.0)	1 (0.8)	<0.001 (<.001 to >999.999)	0 (0.7)	1 (0.8)	NA
Smoking + hazards	2 (5.4)	3 (2.5)	8.33 (0.38-182.89)	3 (4.2)	5 (2.5)	9.05 (0.54-152.17)
Smoking + hazards + bed sharing	15 (40.5)	1 (0.8)	514.26 (17.22 to >999.999)	18 (27.4)	1 (0.7)	450.61 (26.84-7593.14)
None	4 (10.8)	63 (52.9)	Reference	6 (10.1)	76 (54.7)	Reference

in a cot or cot-type bed (adult bed: unimputed unadjusted OR 7.14, 95% CI 4.33-11.75; and sofa/other OR 7.12, 95% CI 3.15-16.08). Pillows were more likely to be used by case infants compared with controls during the last or nominated sleeps (n = 72, 69.3%; n = 77, 30.4%, respectively). A univariable analysis based on unimputed data on the effect of pillows in the bed but not under the infant, identified an increased risk of SUDI (OR 3.83, 95% CI 2.06-7.11). The risk further increased (OR 6.55, 95% CI 3.69-11.62) when pillows were positioned underneath the infant (Table V). The degree to

which infants were tucked into their beds, whether in cots, adult beds, or sofas, was associated with SUDI risk. Being firmly or very firmly tucked compared with not tucked was protective (OR 0.24, 95% CI 0.13-0.44) (Table V).

Mattress Softness

Mattress softness measurements indicated very little difference in the softness of sleep surfaces of cases and controls between being placed to sleep and being found unresponsive (cases) or awake (controls). However, increased mattress

Table V. Bed used for last (cases) and nominated (controls) sleep

Place of last (cases) or nominated (controls) sleep	Cases (%) n = 132	Controls (%) n = 258	Unimputed univariable OR (95% CI)
Bed sharing	(Missing = 6)	(Missing = 0)	$P \leq .0001$
No	53 (42.1)	212 (82.2)	Reference
Yes	73 (57.9)	46 (17.8)	6.35 (3.94-10.22)
Bed used for last (cases) or nominated (controls) sleep	(Missing = 5)	(Missing = 0)	$P \leq .0001$
Cot/cot-type bed*	39 (30.7)	196 (76.0)	Reference
Adult bed	71 (55.9)	50 (19.4)	7.14 (4.33-11.75)
Sofa/other†	17 (13.4)	12 (4.7)	7.12 (3.15-16.08)
Pillows used for last (cases) or nominated (controls) sleep	(Missing = 28)	(Missing = 4)	$P \leq .0001$
No pillows	32 (30.8)	177 (69.7)	Reference
Yes, under baby	45 (43.3)	38 (15.0)	6.55 (3.69-11.62)
Yes, but not under baby	27 (26.0)	39 (15.4)	3.83 (2.06-7.11)
How firmly baby was tucked in when placed to sleep	(Missing = 29)	(Missing = 6)	$P \leq .0001$
Not tucked (including no bedding over baby) or loosely tucked	64 (62.1)	97 (38.5)	Reference
Neither loosely nor firmly tucked	23 (22.3)	52 (20.6)	0.67 (0.37-1.20)
Firmly or very firmly tucked	16 (15.5)	103 (40.9)	0.24 (0.13-0.44)
Sub-section	(Missing = 93)	(Missing = 62)	
Cot (or similar) used for last (cases) or nominated (controls) sleep			
Cot bumpers or similar in the cot	(Missing = 105)	(Missing = 71)	$P = .29$
Yes	3 (11.1)	37 (19.8)	0.51 (0.15-1.77)
No	24 (88.9)	150 (80.2)	Reference
Soft toys in the cot	(Available n = 23)	n = 186)	$P = .68$
Yes	7 (30.4)	49 (26.3)	1.22 (0.48-3.15)
No	16 (69.6)	137 (73.7)	Reference
Objective measures of mattress softness	(Missing = 77)	(Missing = 59)	
Mattress softness where placed	23.3 mm (SD 5.9)	21.2 mm (SD 6.6)	$P = .03$
Mattress softness where found/woke	23.0 mm (SD 6.3)	21.3 mm (SD 6.7)	$P = .09$
			1.04 (0.99-1.09)

*Cot-type beds—controls: Pēpi-Pod × 6, Moses basket × 8, hammock × 2, and wahakura × 1; cases: Pepi-Pod × 1, hand-made or household item × 1.

†Other—controls: pushchair × 2, held/cradled × 2, car × 1, sofa-bed × 1, and bouncer × 1; cases: nil.

softness was seen among cases and controls (23.3 mm vs 21.2 mm) when placed and showed a statistically significant relationship to SUDI risk (unimputed aOR 1.05 per mm, 95% CI 1.00, 1.10; $P = .03$) (Table V). This equates to an increased risk of 1.63 per centimeter. Based on the difference between cases and controls of 2.1 mm, the increase in risk is approximately 1.11 for the average case, compared with the average control. The large SD indicates that some infants were at a marked increased risk due to mattress softness.

Cot Bumpers and Toys

Cot bumpers were used by a small proportion of infants (cases: $n = 3$, 11.1%; controls: $n = 37$, 19.8%) and did not present a significant increased risk of SUDI (OR 0.51, 95% CI 0.15-1.77). Likewise, soft toys in the cot were present in 7 (30.4%) cases and 49 (26.3%) controls and did not show a significant risk (OR 1.22, 95% CI 0.48-3.15) (Table V).

Discussion

We have shown that the prevalence of hazardous situations, in which infants are exposed to sleeping on a sofa, or to caregivers that have recently used alcohol and/or cannabis and other drugs (controls <0.5%, 7.2%, and <0.5%, respectively), is low in our high-risk control population. However, in situations where they do exist, the risk to infants appears to be very high, particularly in combination with maternal smoking and bed sharing.

The size of the infant population at risk from sofa-sleeping is unclear, as few studies have investigated this practice in detail. Infant deaths on sofas have been associated with side sleeping, changes in infant sleep location, and/or bed type by caregivers during the last sleep, exposure to tobacco smoking in pregnancy, and infants sleeping with others on a sofa.⁹ Of the hazardous situations reported in this study, very few infants slept on sofas (controls $n = 1$; cases $n = 11$). The 8 infants who died while sharing the sofa were placed and found during the night and early morning, and they were at statistically significant increased risk of SUDI, which is consistent with previous research in this area.⁷⁻⁹

The effects of habitual alcohol intake on the risk of SUDI are uncertain, as previous studies have found nil or weak evidence.²³ In the present study, recent use of alcohol and/or cannabis and other drugs by caregivers was uncommon in the study population, and individually there was no statistically significant effect on SUDI risk when analysis was carried out using imputed data. Few caregivers in the control group reported using alcohol before or during the nominated sleep (7.2%). In comparison, the Ministry of Health's New Zealand Annual Survey reported that 20% of adults aged 15 years or older drank hazardously in the 12 months to mid-2019.²⁴ Although pregnant women and women of child-bearing age would likely comprise only a small portion of these figures, it may suggest that mothers in the control group were adhering to national guidance to avoid alcohol

during infant care. Alternatively, the knowledge that they were drinking whilst caring for an infant could have led to under reporting.

New Zealand research in the 1990s reported that recent maternal alcohol use was not an independent risk factor for SUDI.²³ An analysis of pooled data from 5 case-control studies—of which a large proportion of the alcohol data was imputed—concluded that infants of all ages of mothers who consumed 2 or more units of alcohol, room-shared, and had a nonsmoking partner, but did not bed share or use drugs or have any other risk factors, were at an approximate 5-fold increased risk of SUDI.²⁵

In our multivariable analyses, the imputed data showed no statistically significant increased risk for recent use by caregivers of alcohol (IaOR 2.40, 95% CI 0.90-6.39 vs aOR 3.04, 95% CI 1.04-8.89) or cannabis and other drugs (IaOR 6.92, 95% CI 0.59-81.02 vs aOR 6.41, 95% CI 0.44-94.26). In the 2012/2013 New Zealand-wide health survey, 8% of women aged 15 years and older reported using cannabis in the previous 12 months.²⁶ As with alcohol use, this could indicate that mothers in the control group, who were generally the main caregivers, followed general health advice not to use cannabis while caring for their infant. Previous New Zealand-based and international research has reported regular cannabis use as a weak risk factor for SUDI,²⁷ and that maternal cannabis use during pregnancy was not associated with an increased risk of SUDI after controlling for tobacco smoking in pregnancy.¹⁰

The prevalence of the hazards (sofa-sleeping, caregiver use of alcohol, and/or cannabis and other drugs) was low in this study. However, the presence of multiple hazards increased the risk of SUDI, especially for very young infants (younger than 3 months of age). The key driver of risk in the interactions was tobacco smoking in pregnancy, whether in combination with bed sharing, or one or more hazards, or both. No other individual factor or combination was significant without smoking.

It should be noted that for young infants, bed sharing without exposure to tobacco smoking or hazards was associated with a statistically significant increased risk of SUDI. This in contrast to the contention of others that it is the presence of smoking or hazards that cause the increased risk associated with bed sharing.

The CIs for the interactions of the combined hazards, bed sharing, and tobacco smoking in pregnancy are wide, due to the very low prevalence of hazards in the control population. This suggests that the missing data were associated with smoking. This is feasible, as the missing data were predominantly from the case group, where Māori were over-represented, and Māori are known to have greater smoking rates than non-Māori.²²

The sleep environment is important as most SUDI deaths happen during sleep, and particularly overnight.^{28,29} Detailed guidelines exist around the safety or otherwise of bed clothes, pillows, and other items in the sleep environment.²⁹ Research has been carried out in relation to thermoregulation,^{29,30} but

few studies have investigated the physical nature of the sleep environment, including the surface, bedding, and items such as soft toys.

Cots or similar were the most common type of bed used among the controls and remain the safest place for infants to sleep. Pillows in the infant sleep environment have previously been reported as a danger to infants,^{31,32} as has the use of soft bedding during bed sharing.³³ This analysis further adds to this evidence and suggests an increased risk when pillows are in the sleep environment, and a greater risk when under the infant. No risk was observed in this study in relation to cot bumpers or toys in the cot.

The degree to which infants were tucked in, whether in a cot, adult bed, or sofa, was associated with SUDI risk. Being firmly or very firmly tucked in was protective (OR 0.24, 95% CI 0.13-0.44), compared with being untucked, loosely tucked, or uncovered (Table V). These findings echo those of the New Zealand Cot Death Study (1989-1991), in which firm tucking contributed to reducing the risk of SUDI.³¹

There was no discernible difference in SUDI risk among Māori and non-Māori controls in relation to use of a sofa as the nominated sleep space and/or recent use by caregivers of alcohol and/or cannabis and other drugs. This suggests that our previous findings, in which smoking in pregnancy is the key difference in the disparity in SUDI experienced by Māori compared with non-Māori, are still accurate.²² We note that maternal smoking of tobacco during pregnancy and after the birth of the child is highly correlated, but the effect of smoking in utero on the fetus is profound, resulting in reduced birthweight and reduced arousal response.³⁴⁻³⁷

Key strengths of the study were the inclusion of nearly all eligible cases (96%), and the collection of complete datasets from controls. Limitations included a lower-than-expected response rate for controls and missing data across the variables for cases, compared with controls, although this has been mitigated by imputation. Imputation helped to minimize the issue of missing data, but it is also a limitation. However, a sensitivity analysis of the original individual participant data meta-analysis of SIDS and bed sharing showed that the results were consistent with the imputed results, suggesting we can have confidence in the imputed results.³⁸

The combination of tobacco smoking in pregnancy and bed sharing continues to present the greatest SUDI risk for infants, and very young infants are at even greater risk, which further increases with each additional hazard (sofa-sleeping and/or recent use by caregivers of alcohol and/or cannabis and other drugs). For infants younger than 3 months, bed sharing even without smoking and hazards is associated with an increased risk of SUDI. Further initiatives are required to address the environment in which a large proportion of SUDI deaths occur. Given that smoking remains a pivotal and modifiable risk factor, there is continued need for the implementation of effective, appropriate programs for smoking cessation, safe sleep, and the supply of safe sleep beds, to reduce SUDI rates in New Zealand and address the significant and ongoing SUDI disparity among Māori and other vulnerable populations. ■

We extend our sincere thanks to the following people and organizations: the immediate past and present Chief Coroners of New Zealand, Judge Neil MacLean and Judge Deborah Marshall, and the Ministry of Justice, including the National Initial Investigation Office and the coroners; members of the SUDI Study Steering Committee; and to previous organizations, Whakawhetū (Māori SUDI Prevention) and Taha (Pacific SUDI Prevention).

Special thanks to the SUDI Liaison Team Trainer, Ms Yvonne Ledesma, Lead SUID Investigator, Miami-Dade Medical Examiner's Office, Florida, USA; and Mr David Aro, Communio Consultancy, for implementing the study; and the past and present members of the national SUDI Liaison Team, especially Ms Shelley Jonas and Ms Elaine McLardy. Above all, heartfelt thanks to the infants and families who participated in the study or have been affected by SUDI.

This research was conducted during the Lead Author's tenure of a 'Māori Health PhD Scholarship' of the Health Research Council of New Zealand.

Submitted for publication May 3, 2021; last revision received Jan 20, 2022; accepted Jan 26, 2022.

Reprint requests: M. MacFarlane, Department of Paediatrics: Child and Youth Health, University of Auckland, Private Bag 92019, Auckland 1142, New Zealand. E-mail: m.macfarlane@auckland.ac.nz

References

- Willinger M, James LS, Catz C. Defining the sudden infant death syndrome (SIDS): deliberations of an expert panel convened by the National Institute of Child Health and Human Development. *Pediatr Pathol* 1991;11:677-84.
- Taylor BJ, Garstang J, Engelberts A, Obonai T, Cote A, Freemantle J, et al. International comparison of sudden unexpected death in infancy rates using a newly proposed set of cause-of-death codes. *Arch Dis Child* 2015;100:1018-23.
- Byard RW. Sudden unexpected death in infancy (SUDI)—the role of the pathologist. *Curr Pediatr Rev* 2010;6:21-6.
- Mitchell E. Co-sleeping and suffocation. *Forensic Sci Med Pathol* 2015;11:227-78.
- Mitchell EA, Thompson JM, Zuccollo J, MacFarlane M, Taylor B, Elder D, et al. The combination of bed sharing and maternal smoking leads to a greatly increased risk of sudden unexpected death in infancy: the New Zealand SUDI Nationwide Case Control Study. *N Z Med J* 2017;130:52-64.
- Escott A, Elder DE, Zuccollo JM. Sudden unexpected infant death and bedsharing: referrals to the Wellington Coroner 1997-2006. *N Z Med J* 2009;122:59-68.
- Blair PS, Sidebotham P, Pease A, Fleming PJ. Bed-sharing in the absence of hazardous circumstances: is there a risk of sudden infant death syndrome? An analysis from two case-control studies conducted in the UK. *PLoS One* 2014;9:e107799.
- Byard RW, Beal S, Blackbourne B, Nadeau JM, Krous HF. Specific dangers associated with infants sleeping on sofas. *J Paediatr Child Health* 2001;37:476-8.
- Rechtman LR, Colvin JD, Blair PS, Moon RY. Sofas and infant mortality. *Pediatrics* 2014;134:e1293-300.
- Klonoff-Cohen H, Lam-Kruglick P. Maternal and paternal recreational drug use and sudden infant death syndrome. *Arch Pediatr Adolesc Med* 2001;155:765-70.
- El Marroun H, Brown QL, Lund IO, Coleman-Cowger VH, Loree AM, Chawla D, et al. An epidemiological, developmental and clinical overview of cannabis use during pregnancy. *Prev Med* 2018;116:1-5.
- Jaques SC, Kingsbury A, Henshcke P, Chomchai C, Clews S, Falconer J, et al. Cannabis, the pregnant woman and her child: weeding out the myths. *J Perinatol* 2014;34:417-24.
- Poutasi K. Ethnicity data protocols for the health and disability sector. Wellington, New Zealand: Ministry of Health; 2004.
- Alcohol [Internet]. 2019. Accessed August 12, 2020. <https://www.health.govt.nz/your-health/healthy-living/addictions/alcohol-and-drug-abuse/alcohol>

15. Tipene-Leach D, Abel S. The wahakura and the safe sleeping environment. *J Prim Health Care* 2010;2:81.
16. Cowan S. The Pēpi-Pod Programme 2013 Report. Christchurch: Change for our Children Limited; 2014.
17. Schlaud M, Dreier M, Debertin AS, Jachau K, Heide S, Giebe B, et al. The German case-control scene investigation study on SIDS: epidemiological approach and main results. *Int J Legal Med* 2010;124:19.
18. Child and Youth Mortality Review Committee, Te Roopu Arotake Auau Mate o te Hunga Tamariki, Taiohi. Fifth Report to the Minister of Health: Reporting mortality 2002-2008. Wellington: Child and Youth Mortality Review Committee; 2009. Report No.: 5.
19. Mitchell EA, Cowan S, Tipene-Leach D. The recent fall in post-perinatal mortality in New Zealand and the Safe Sleep Programme. *Acta Paediatr* 2016;105:1312-20.
20. SAS Institute Inc. SAS/STAT 15.1 user's guide. Cary (NC): SAS Institute Inc.; 2018. p. 6376.
21. Live Births, Stillbirths by District Health Board (Maori and Live Births) [Internet]. 2016. Accessed April 20, 2017. http://www.stats.govt.nz/browse_for_stats/population/births/births-tables.aspx
22. MacFarlane M, Thompson J, Zuccollo J, McDonald G, Elder D, Stewart AW, et al. Smoking in pregnancy is a key factor for sudden infant death among Māori. *Acta Paediatr* 2018;107:1924-31.
23. Scragg R, Mitchell EA, Taylor BJ, Stewart AW, Ford RP, Thompson JM, et al. Bed sharing, smoking, and alcohol in the sudden infant death syndrome. New Zealand Cot Death Study Group. *BMJ* 1993;307:1312-8.
24. New Zealand Health Survey: Annual Data Explorer, November 2019 [Internet]. 2019. Accessed August 7, 2020. https://minhealthnz.shinyapps.io/nz-health-survey-2018-19-annual-data-explorer/_w_337a677b/#!/explore-topics
25. Carpenter R, McGarvey C, Mitchell EA, Tappin DM, Vennemann MM, Smuk M, et al. Bed sharing when parents do not smoke: is there a risk of SIDS? An individual level analysis of five major case-control studies. *BMJ Open* 2013;3:e002299.
26. Ministry of Health. Cannabis Use 2012/13: New Zealand Health Survey. Wellington: Ministry of Health; 2015.
27. Scragg R, Mitchell EA, Ford R, Thompson J, Taylor BJ, Stewart AW. Maternal cannabis use in the sudden death syndrome. *Acta Paediatr* 2001;90:57-60.
28. Leach CE, Blair PS, Fleming PJ, Smith IJ, Platt MW, Berry PJ, et al. Epidemiology of SIDS and explained sudden infant deaths. *Pediatrics* 1999;104:e43.
29. Moon RY. Task Force on Sudden Infant Death Syndrome. SIDS and other sleep-related infant deaths: evidence base for 2016 updated recommendations for a safe infant sleeping environment. *Pediatrics* 2016;138:e20162940.
30. Galland BC, Elder DE. Sudden unexpected death in infancy: biological mechanisms. *Paediatr Respir Rev* 2014;15:287-92.
31. Wilson CA, Taylor BJ, Laing RM, Williams SM, Mitchell EA. New Zealand Cot Death Study Group. Clothing and bedding and its relevance to sudden infant death syndrome: further results from the New Zealand Cot Death Study. *J Paediatr Child Health* 1994;30:506-12.
32. Blair PS, Sidebotham P, Evason-Coombe C, Edmonds M, Heckstall-Smith EM, Fleming P. Hazardous cosleeping environments and risk factors amenable to change: case-control study of SIDS in south west England. *BMJ* 2009;339:b3666.
33. Blair PS, Ball HL, McKenna JJ, Feldman-Winter L, Marinelli KA, Bartick MC, et al. Bedsharing and Breastfeeding: The Academy of Breastfeeding Medicine Protocol# 6, Revision 2019. *Breastfeed Med* 2020;15:5-16.
34. Abraham M, Alramadhan S, Iniguez C, Duijts L, Jaddoe VW, Den Dekker HT, et al. A systematic review of maternal smoking during pregnancy and fetal measurements with meta-analysis. *PLoS One* 2017;12:e0170946.
35. Juárez SP, Merlo J. Revisiting the effect of maternal smoking during pregnancy on offspring birthweight: a quasi-experimental sibling analysis in Sweden. *PLoS One* 2013;8:e61734.
36. Tirosh E, Libon D, Bader D. The effect of maternal smoking during pregnancy on sleep respiratory and arousal patterns in neonates. *J Perinatol* 1996;16:435-8.
37. Richardson HL, Walker AM, Horne RS. Maternal smoking impairs arousal patterns in sleeping infants. *Sleep* 2009;32:515-21.
38. Carpenter JR, Smuk M. Missing data: a statistical framework for practice. *Biom J* 2021;63:915-47.