

## **GENERAL PRACTITIONERS' FAMILIARITY WITH SUDDEN INFANT DEATH SYNDROME IN THE UK: THE RESULTS OF THE SIDS PROJECT**

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**Abstract.** *The purpose of this study is to provide information about General Practitioners' (GPs) knowledge and behaviour about Sudden Infant Death Syndrome (SIDS) and its risk factors in the United Kingdom. We investigated if GPs knew that the supine sleep position is best for reducing the risk of SIDS. We also explored their overall knowledge regarding all SIDS risk factors and their recommendations to parents. Approximately 13% of GPs did not give the correct answer about the safest sleep position. Being female, young age, having children, the number of practices where the GP works and direct experience of a case of SIDS resulted in being the strongest determinants of knowledge. On the contrary, a post-graduate title in child health and paediatrics did not significantly increase GPs's knowledge of SIDS. Significant differences among regions emerged and were likely to be the result of training and prevention campaigns undertaken in these regions.*

**Keywords:** *Sudden Infant Death Syndrome; general practitioners; infant mortality; knowledge assessment; sleep position.*

### **1. INTRODUCTION**

Sudden Infant Death Syndrome (SIDS, also known as 'cot death' or 'crib death') is defined as 'the sudden unexpected death of an infant <1 year of age, with onset of the fatal episode apparently occurring during sleep, that remains unexplained after a thorough investigation, including performance of a complete autopsy and review of the circumstances of death and the clinical history' (Krous et al., 2004). Nowadays, SIDS is still the major cause of death in healthy infants born in

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developed countries, with an incidence rate which varies between 0.08 and 0.43 deaths per 1,000 infants (Centraal Bureau voor de Statistiek, 2013; Hoyert and Xu, 2012). However, the cause of SIDS is still unclear, so it is not possible to take action that will definitely prevent it (Mitchell, 2009).

Over the years many epidemiological studies discovered a number of behaviours which can significantly affect the risk of SIDS, making it possible to implement some simple interventions in order to reduce it. The sleep position is the strongest risk factor on which it is possible to intervene in order to reduce the risk of SIDS, and among all positions the supine one is the safest. The American Academy of Pediatrics (AAP) considers as 'A-level recommendations' for reducing the risk of SIDS those presented in Table 1 (Task Force on SIDS, 2011); however, it is important to remember that each country has its own specific recommendations.

**Table 1: 'A-level recommendations' for the prevention of SIDS (Task Force on SIDS 2011)**

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1. To put the infant to sleep supine on a firm surface and in an environment free of soft objects and loose bedding;
  2. To avoid overheating of the infant's room;
  3. To give infants a pacifier before putting them to sleep;
  4. To share the same room with the infants but not the bed;
  5. To breastfeed;
  6. To receive proper prenatal care for pregnant women;
  7. To avoid smoking, alcohol and drugs consumption during and after pregnancy;
  8. To avoid the use of home cardiorespiratory monitors as a strategy for reducing the risk of SIDS;
  9. To actively involve paediatricians, family physicians and other primary care professionals in the campaigns focused on preventing SIDS.
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This paper reports on the results of a survey carried out by the SIDS Project, a research study meant to provide the first data about the awareness and behaviour of General Practitioners (GPs) regarding SIDS and its risk factors in the United Kingdom. However, due to budget constraints, it could only be carried out in the region corresponding to the South Central Strategic Health Authority (which includes the counties of Berkshire, Buckinghamshire, Hampshire, Isle of Wight, and Oxfordshire). All the GPs working in the relative area were chosen as the target population. GPs were chosen considering their long-term relationship of trust and confidence with the infants' parents and because parents often refer to their GP to seek advice and recommendations for issues about their infants. We are aware that

midwives and health visitors could also have been included in the target population; however, these populations would have been very different from the GPs and we would have needed more time and funding than those available, so we preferred to leave such a possibility to an eventual future research project.

The first aim of this work is to analyse GPs' knowledge about SIDS most important risk factor, which is the sleep position. In order to do so, we investigated if GPs knew that the supine sleep position alone is the best to reduce the risk of SIDS and we described those who knew it and those who did not on the basis of their demographic and professional background.

Secondly, considering that GPs may discuss also all the other SIDS risk factors with parents, we wanted to verify what was the GPs' overall level of knowledge of all SIDS risk factors. Once again, this analysis was carried out on the basis of their demographic and professional background.

Finally we analysed GPs' recommendations about infant sleep position. We investigated if they recommended exclusively the supine sleep position and identified those who recommended it and those who did not.

## **2. THE SURVEY**

The study consisted of a mail survey carried out in the United Kingdom between May and July 2012. The survey design was cross-sectional and it included three different mailings. Two weeks passed between each of the mailings. The sample frame was retrieved from the website of the National Health Service (NHS). A previously validated questionnaire was updated with additional details (de Luca and Boccuzzo, 2014). The variables of interest consisted of one question that asked respondents which was the safest sleep position, 14 questions about SIDS risk factors and one question regarding recommendations about the sleep position and their frequency. The possible answers to the questions about the safest sleep position and the recommendations given to parents were 'Supine', 'Lateral', 'Prone', 'I do not know'. Demographic variables were also included. Response to the survey was considered as consent to participate. The SIDS project was approved by the Ethics Committee and the Research Governance Office of the University of Southampton (Project ID: 1197).

The variables of interest consisted of all the items that described healthcare professionals' knowledge and recommendations about SIDS and its risk factors. As these were dichotomous (correct/non correct), we imposed a level of precision of 0.05, a confidence level of 95%, and a distribution of the answers in the population of 50% ( $p=0.5$ ) in order to determine the sample size. The choice of  $p=0.5$  was made

because we thought that in some cases the distribution of the answers in the population could well have been of 50%. As a result, the required sample size for a target population of 2,658 GPs was 336. In order to estimate the expected response rate we considered 28 surveys previously carried out on this topic (Table 2). Using the information given by all of them, the resulting expected response rate would have been 64.3%. However, this survey was bigger than most of them, so we preferred to focus only on those studies with a sample size of at least 1,000 participants. After adjusting for the effects of reminders (a +10% effect for each of the two reminders – Dillman et al., 2009), the expected response rate was 20.7%, which was rounded to 21%. As a consequence, with an initial response rate of 21% and a +10% effect for each of the two reminders, the overall sample size for the survey was of 820. This quantity eventually became 823 after stratifying (with proportional allocation of stratum sample size) for gender and average size of the practice where the GP works.

**Table 2: Estimation of the expected response rate**

	<b>All previous surveys (unadjusted)</b>	<b>Surveys with samples &gt; 1,000 (unadjusted)</b>	<b>Surveys with samples &gt; 1,000 (adjusted)</b>
Number of surveys	28	5	5
Average sample size	522	1,713	1,713
Median response rate	68.6%	31.6%	21.6%
Average response rate	64.3%	27.7%	20.7%

The overall response rate was 42.4% (349 responses), which is one of the highest response rates registered for studies on this topic (de Luca and Hinde, 2016). There were no statistically significant differences in response rates by county (Table 3). There were slightly more females (n = 180, 51.6%) than males, and the majority of GPs (85.5%) obtained their degree in the United Kingdom. Other personal and demographic information is shown in Table 3.

**Table 3: Descriptive statistics of the sample**

Variable	Categories	% (if not otherwise stated)
Gender	Male	48.4
Children	No children	11.6
	Aged 0-2	15.6
	Aged 3 or more	72.5
Citizenship	UK	93.7
Country of medical degree	UK	88.5
Number of practices where the GP works	One	84.5
County	Berkshire	16.9
	Buckinghamshire	13.5
	Hampshire	35.2
	Oxfordshire	18.3
	Isle of Wight <sup>a</sup>	16.1
Number of inhabitants in the city of GP practice	< 10k	28.2
	10k ≤ & < 20k	15.2
	20k ≤ & < 40k	14.4
	40k ≤ & < 100k	18.7
	≥ 100k	23.6
GP with more female colleagues than males		41.0
GP did a placement in child health and paediatrics		76.2
GP holds a post-graduate degree in child health and paediatrics		26.4
GP has direct experience of a case of SIDS		45.1
Average age (SE)		47.5 (0.46)
Average years of experience (SE)		16.6 (0.48)
Average number of colleagues in the workplace (SE)		6.5 (0.14)

<sup>a</sup> The cities of Portsmouth and Southampton were included in the Isle of Wight County to balance the county sample sizes.

### 3. STEPS OF THE ANALYSIS

#### 3.1 STEP 1: THE AWARENESS OF THE SAFEST SLEEP POSITION

The first step of the analysis consisted in identifying the factors that influenced GPs' awareness of the safest sleep position. As the nature of the target variable was hypothesised dichotomous, we adapted a log binomial model to the data (Wacholder, 1986). This model belongs to the Generalized Linear Models family which is characterized by a logarithmic link function and a binomial distribution:

$$\Pr(Y_i = 1 | x_i) = e^{x_i' \beta} \quad (1)$$

where:

$Y_i$  indicates the dichotomous random outcome for the  $i$ -th respondent;

$x_i' = \{x_{i1}, \dots, x_{is}\}$  indicates the values of a set of  $S$  covariates for the  $i$ -th respondent;

$\beta = \{\beta_0, \beta_1, \dots, \beta_s\}$  indicates the  $(S+1)$  regression parameters.

The choice of this model was made after realizing that the number of GPs who did not know that the supine position was the safest for SIDS prevention was not small. In fact, out of 349 respondents, 46 (13.2%) did not give the correct answer. This number was too high to rely on the usual approximation of the risk ratios given by the odds ratios, so we could not use a logistic regression in order to identify the risk factors of respondents who did not reply correctly.

The biggest shortfall of the log binomial model is represented by his high 'failure' rate (Blizzard and Hosmer, 2006), mainly due to 1) predicted probabilities that are not bounded between 0 and 1 (this is a consequence of using a logarithmic link function instead of a logit); and 2) computational issues that drive to the non-convergence of the model. In our case, however, neither problem was detected.

#### 3.2 STEP 2: AN INDEX OF GENERAL KNOWLEDGE

In order to build an index of how much GPs actually knew about SIDS and its risk factors, we considered 14 items that were included under the same question: 'What effects do you believe that the following behaviours have on the risk of SIDS?'. Respondents could choose between 'it lowers the risk', 'it increases the risk', 'it does not affect the risk' and 'I do not know'. The items and the correct answers (based on the advice of the AAP, Task Force on SIDS 2011) are presented in Table 4.

**Table 4: Potential risk factors and related effects considered in the questionnaire**

Risk factor	Effect
Placing infants to sleep in a supine position	It lowers the risk
Offering infants a dummy at nap time and bedtime	It lowers the risk
Using a soft crib mattress	It increases the risk
Allowing infants to sleep in the same bed as their parents	It increases the risk
Encouraging tummy time when the infant is awake and observed	It does not affect the risk
Making up the bedding so that the infant's feet reach the foot of the crib	It does not affect the risk
Performing an electrocardiogram on the infant	It does not affect the risk
Keeping the bedroom temperature below 20° C	It lowers the risk
Maternal smoking during pregnancy	It increases the risk
Allowing infants to sleep in the same room as their parents	It lowers the risk
Breastfeeding	It lowers the risk
Placing soft objects such as pillows, quilts and stuffed toys in the crib	It increases the risk
Smoking (both maternal and paternal) in the infant's environment	It increases the risk
Sleeping with an infant on a couch / armchair	It increases the risk

In order to obtain the index of knowledge, we adapted to the data a Rasch model (Rasch, 1960; Wright, 1977; Wright and Masters, 1982; Fisher, 1995). The Rasch model, similarly to the other models belonging to the Item Response Theory family, allows translating variables measured on a metric scale into scores that refer to a latent continuum (in this case the latent variable was given by the knowledge of respondents). The formulation of the Rasch model that has been used for the construction of the index is the two-parameter Rasch model for dichotomous responses (Birnbaum, 1968):

$$\Pr(x_{in} = 1|\theta_n) = \frac{\exp\{\lambda_i(\theta_n - \delta_i)\}}{1 + \exp\{\lambda_i(\theta_n - \delta_i)\}} \quad (2)$$

where:

$X_{in} = 0, 1$  is the variable that describes the answer given by the  $n$ -th respondent to the  $i$ -th item (where 0 represents an incorrect answer, and 1 a correct answer);

$\theta_n$  indicates the knowledge of the  $n$ -th respondent: the greater this parameter, the greater the probability that the respondent would give the correct answer;

$\delta_{il}$  indicates the difficulty parameter associated with the transition from 0 to 1 for the  $i$ -th item. The greater this parameter, the greater the probability that the respondent would give an incorrect answer (0). The estimate of  $\delta_{il}$  represents the value at which an individual with an ability parameter  $\theta_n$  equal to  $\delta_{il}$  will have a probability of giving a correct answer to the  $i$ -th item of 0.5. The higher the difference between  $\theta_n$  and  $\delta_i$ , the higher the probability that the respondent will give a correct answer;

$\lambda_i$  indicates the discrimination parameter for the  $i$ -th item. The parameter  $\lambda_1$ , the one referring to the first item, is set equal to 1 because of identification; as a consequence, the first item acts as the item of reference for all the other items. The degree to which  $\lambda_i$  is greater than 1 indicates the stronger discrimination power of the  $i$ -th item with respect to the item of reference, while the degree to which  $\lambda_i$  is less than 1 indicates the weaker degree of discrimination power of the  $i$ -th item with respect to the item of reference. An item with  $\lambda_i$  greater than 1, then, will be better in distinguishing between more and less knowledgeable respondents than the reference item. An item with  $\lambda_i$  less than 1, instead, will be less effective.

While the original Rasch model does not consider the discrimination parameter ( $\lambda_i$ ), we believed that this parameter deserved to be taken into account. In fact, it could not be assumed that all items had an equal effectiveness in discriminating knowledgeable respondents. The opportunity of including it in the model was tested with a simple likelihood-ratio test between the model with  $\lambda_i$  and the one without it. The inclusion of the parameter was then supported by the result of this test ( $\chi^2_{13} = 75.0, p < 0.001$ ).

The correct application of the Rasch model is constrained to the fundamental assumption of one-dimensionality; that is, to the assumption that the  $I$  items being used are all indicators of the latent variable of interest. With this goal in mind, we ran a factor analysis, and its results showed that a high proportion of variance was explained by the first dimension (73.5%).

One of our main objectives was to investigate which demographic and professional characteristics significantly affected GPs' knowledge about SIDS and its risk factors. To this end, the index of knowledge  $\theta_n$  is our dependent variable.

To model the index of knowledge we used the quantile regression approach (Koenker, 2005). This choice was justified by several reasons. First of all, we were particularly interested in analysing the determinants of knowledge at the extreme values of the index. In other words, we were interested in investigating the characteristics of the less and better prepared respondents. To do so, we



decided to focus also on the 20<sup>th</sup> and 80<sup>th</sup> percentiles of the distribution rather than only to the median. More generally, the quantile regression was chosen because it enabled us to understand if the determinants of GPs' knowledge changed at different levels of the dependent variable.

The linear quantile regression model is:

$$y_i = \mathbf{x}'_i \boldsymbol{\beta}_p + \varepsilon_i \quad (3)$$

where:

$y_i$  indicates the continuous outcome for the  $i$ -th respondent;

$\mathbf{x}'_i = \{x_{i1}, \dots, x_{iS}, \dots, x_{iS}\}$  indicates the values of a set of  $S$  covariates for the  $i$ -th respondent;

$\boldsymbol{\beta}_p = \{\beta_{p0}, \beta_{p1}, \dots, \beta_{pS}, \dots, \beta_{pS}\}$  indicates the  $(S+1)$  regression parameters for  $p$ -th quantile.

As a consequence, the  $p$ -th quantile is given by:

$$p = P(y_i \leq \mathbf{x}'_i \boldsymbol{\beta}_p | \mathbf{x}_i) \quad (4)$$

and the  $p$ -th quantile of the conditional distribution of  $y_i$  given  $\mathbf{x}_i$  is:

$$Q_y(p) = \mathbf{x}'_i \boldsymbol{\beta}_p . \quad (5)$$

The quantile regression can be applied to the data regardless of the distribution of the variable of interest, thus making unnecessary any hypothesis about it. The interpretation of the  $\beta_{pS}$  parameter is similar to the one in a simple linear model.  $\beta_{pS}$  represents the change of the  $p$ -th quantile of  $y$  in correspondence to a unitary change of  $x_s$ , while all the other explanatory variables remain constant (Koenker, 2005).

### 3.3 STEP 3: CHARACTERISTICS OF THE GPs GIVING RECOMMENDATIONS TO PARENTS

In order to understand the characteristics of the GPs who talked with parents about SIDS and gave them recommendations about the sleep position, we modelled both the variable  $S$ ="GP talks with parents" and, among those GPs that talked with parents about the sleep position, the variable  $Y$ ="GP gives the correct recommendation". This was necessary because not all GPs talked with parents about the sleep position, and hence  $Y$  was observed only if a selection condition was met. In such circumstances, modelling two independent equations with standard regression techniques would have resulted in biased and inconsistent estimators if unobserved factors affecting  $Y$  were correlated with unobserved factors affecting the selection process  $S$  (Heckman, 1979). However, contrarily to the classical Heckman's sample selection model, in our case the variable of interest  $Y$  was binary

and not continuous. In order to account for this difference, we applied a model formulated as a system of equations for two latent responses,  $y_i^*$  and  $S_i^*$  (Miranda and Rabe-Hesketh, 2006):

$$y_i^* = \mathbf{x}_i' \boldsymbol{\beta} + u_i \quad (6)$$

$$S_i^* = \mathbf{z}_i' \boldsymbol{\gamma} + v_i . \quad (7)$$

In this model:

$y_i^*$  and  $S_i^*$  are latent continuous variables;

$\mathbf{x}_i$  (of dimensions  $K \times 1$ ) and  $\mathbf{z}_i$  ( $L \times 1$ ) are vectors of explanatory variables;

$\boldsymbol{\beta}$  ( $K \times 1$ ) and  $\boldsymbol{\gamma}$  ( $L \times 1$ ) are vectors of parameters to be estimated.

The observed responses are generated as:

$$y_i = \begin{cases} 1 & \text{if } y_i^* > 0 \\ 0 & \text{otherwise} \end{cases} \quad (8)$$

$$S_i = \begin{cases} 1 & \text{if } S_i^* > 0 \\ 0 & \text{otherwise} . \end{cases} \quad (9)$$

A bivariate normal distribution is assumed for  $u_i$  and  $v_i$ . A shared random effect  $\varepsilon_i$  is used to induce the dependence between  $u_i$  and  $v_i$ :

$$u_i = \lambda \varepsilon_i + \tau_i \quad (10)$$

$$v_i = \varepsilon_i + \zeta_i \quad (11)$$

where:

$\varepsilon_i$ ,  $\tau_i$  and  $\zeta_i$  are normally distributed with mean 0 and variance 1;

$\lambda$  is a parameter to be estimated.

The correlation between  $u_i$  and  $v_i$  is  $\rho = \frac{\lambda}{\sqrt{2(\lambda^2 + 1)}}$ . If  $\rho = 0$ , consistent estimates

of  $\boldsymbol{\beta}$  and  $\boldsymbol{\gamma}$  are obtained with ordinary probit regression models; if  $\rho \neq 0$ , estimates are inconsistent. Consistent estimators can be obtained by maximum likelihood estimation of a joint probit model of the outcome and selection variable (Miranda and Rabe-Hesketh, 2006), where the log-likelihood is evaluated using adaptive quadrature (Rabe-Hesketh et al., 2002).

The data analysis was performed with Stata (StataCorp, 2011). Descriptive statistics were calculated both for the demographic characteristics of the sample and for the questions of interest. Additionally, we adapted the Heckman's sample selection model by using the 'heckprob' command (Miranda and Rabe-Hesketh, 2006; Chiburis and Lokshin, 2007; De Luca, 2008), the log-binomial regression and quantile regression by using the SAS/STAT<sup>®</sup> software (SAS 2011).

#### 4. RESULTS

##### 4.1 GPS' AWARENESS OF THE SAFEST INFANT SLEEP POSITION

In Table 5, we present the results of the log-binomial regression investigating the characteristics of the respondents who did not choose the supine position as the only safest position. In the table, we included the only variables that significantly influenced the probability of knowing the correct position after a stepwise selection (with inclusion criteria fixed at  $p = 0.05$ ): 'Age' and 'Whether the GP works in only one practice or not'. As it can be seen, older GPs showed a lower likelihood of having a correct knowledge. This may suggest that once they had received their training on SIDS and its risk factors, they did not get any further updates. Alternatively, it could also mean that their interest in this topic decreased over time as they entered midlife. In both cases, this represents a dangerous situation in a field where even the best practice concerning the most important risk factor can change quickly according to the latest scientific evidence.

As for the variable that describes the number of practices where the GPs work, we interpreted it as the effect of the precariousness of their role. As these GPs are not as present in the surgery as a GP who works exclusively there, we hypothesized that they could be less 'exposed' to children's issues. This hypothesis was made considering that mothers may be more inclined to discuss these topics with the GP they have always dealt with rather than with a GP they may not be very familiar with.

**Table 5: Determinants of GPs' correct knowledge about the safest infant sleep position**

Variable	Risk Ratio	Std. Err.	Significance	95% Confidence Interval	
Age (centred)	0.996	0.002	0.060	0.992	1.000
GP works in only one practice	1.217	0.106	0.025	1.026	1.445

##### 4.2 GPS' AWARENESS OF SIDS RISK FACTORS

The distribution of the answers to the different items investigating the effects of 14 different behaviours on the risk of SIDS showed considerably different results (Table 6). For example, the percentage of correct answers to the item 'Smoking in

the infant's environment' was over 90%, while it was just 11% for the item 'Making the infant's feet reach the foot of the crib'. Apart from smoking habits, which are a well-known risk factor for several diseases and causes of death and thus do not require a specific knowledge, the most recognized risk factor for SIDS was the sleeping position. In this case the percentage of wrong answers was only<sup>2</sup> 8%. For almost all the other factors we could observe high percentages of wrong answers. For instance, there were 72.2% for 'Offering infants a dummy at nap time and bedtime' and 55.6% for 'Encouraging tummy time'.

**Table 6: Distribution of answers given by the respondents to the 14 items (percentages)**

	<i>Correct answers</i>	<i>Wrong answers</i>	<i>Non-response</i>
Smoking in the infant's environment	96.5	2.6	0.9
Maternal smoking during pregnancy	96.0	3.1	0.9
Placing soft objects in the crib	74.8	24.4	0.8
Sleeping with an infant on a couch / armchair	68.5	29.5	2.0
Allowing infants to sleep in the same bed as their parents	84.3	14.3	1.4
Offering infants a dummy at nap time and bedtime	26.1	72.2	1.7
Using a soft crib mattress	49.6	47.8	2.6
Placing infants to sleep in a supine position	91.4	8.0	0.6
Keeping the bedroom temperature below 20° C	64.2	33.8	2.0
Breastfeeding	73.1	26.1	0.8
Allowing infants to sleep in the same room as their parents	52.4	45.9	1.7
Performing an electrocardiogram on the infant	69.9	28.7	1.4
Encouraging tummy time	43.3	55.6	1.1
Making the infant's feet reach the foot of the crib	10.9	87.4	1.7

In order to build an index of knowledge which considered all SIDS risk factors, we adapted to the data a Rasch model for dichotomous items and discrimination parameters. The response options which we considered for each item were 'Correct answer' vs. ('Wrong answer' + 'I do not know')

<sup>2</sup> 13.2% represents the percentage of respondents who, when asked which sleeping position is associated with the lowest risk of SIDS, did not reply supine exclusively. They could choose between 'supine', 'lateral', 'prone', any combination of these positions and 'I do not know'. 8% represents the percentage of respondents who, when asked what is the effect of the supine position on the risk of SIDS, stated that placing infants to sleep in a supine position either increased or did not affect the risk.

Table 7 shows the estimates and the significance levels of the discrimination parameters. The item ‘Sleeping supine’ was chosen as the reference item because it has been the most known protective factor against SIDS for many years. For this reason, it should be among those with the highest discrimination power (respondents that give the wrong answer to this item will be more likely to give wrong answers to the others as well). The table reveals that while several items were assigned higher discrimination powers than ‘Sleeping supine’, none of these could be considered as significantly different from it. On the other hand, three items were assigned significantly lower discrimination powers. These items were ‘Performing an electrocardiogram on the infant’, ‘Encouraging tummy time’, and ‘Making the infant’s feet reach the foot of the crib’. This means that answering correctly to any one of these three items implied a lower probability of also answering the other questions correctly. In other words, the preparation of the respondents was not significantly impacted by their answers to these questions, which is consistent with the fact that these items did not describe behaviours with a recognized significant impact on the risk of SIDS.

**Table 7: Rasch model: estimates of discriminatory parameters, standard errors, and Wald test values and their significance for each of the considered SIDS risk factors**

	<b>Discrimination parameter</b>	<b>Standard error</b>	<b>Wald Test Value</b>	<b>Significance (p-value)</b>
Smoking in the infant’s environment	2.94	1.314	1.48	0.140
Maternal smoking during pregnancy	2.74	1.229	1.41	0.157
Placing soft objects in the crib	2.48	0.945	1.56	0.118
Sleeping with an infant on a couch / armchair	2.00	0.743	1.34	0.180
Allowing infants to sleep in the same bed as their parents	1.16	0.453	0.36	0.717
Offering infants a dummy at nap time and bedtime	1.04	0.414	-0.09	0.927
Using a soft crib mattress	1.04	0.418	0.08	0.932
Placing infants to sleep in a supine position	1.00 ( <i>ref.</i> )			
Keeping the bedroom temperature below 20° C	0.96	0.375	-0.12	0.907
Breastfeeding	0.86	0.347	-0.39	0.694
Allowing infants to sleep in the same room as their parents	0.79	0.313	-0.67	0.502
Performing an electrocardiogram on the infant	0.44	0.233	-2.40	<b>0.017</b>
Encouraging tummy time	0.30	0.193	-3.62	<b>0.000</b>
Making the infant’s feet reach the foot of the crib	-0.34	0.273	-4.91	<b>0.000</b>

The index of knowledge  $\theta_n$ , defined as the score attributed by the Rasch model to each respondent, ranged from -2.49 to 1.21. The mean was 0 and the median 0.09; the normality tests rejected the null hypothesis of Gaussian distribution. As we were interested in investigating potential differences in the effect of the explanatory variables according to a specific level of knowledge, we modelled our index with a quantile regression, and we looked at the results for the 20<sup>th</sup>, 50<sup>th</sup> and 80<sup>th</sup> percentiles (Table 8).

**Table 8: Determinants of GPs' correct knowledge about 14 risk factors by percentile (coefficients of the quantile regression)**

Variable	20 <sup>th</sup> percentile	50 <sup>th</sup> percentile	80 <sup>th</sup> percentile
Direct experience of a case of SIDS	-0.2047*	0.2707***	0.1431
Age	-0.0382***	-0.0281***	-0.0277***
Gender (Female)	0.3581**	0.4646***	0.3040***
GP has children	0.3392*	0.4923***	0.4030**
Intercept	0.7362*	0.5914**	1.2680***

\* 0.01 < p < 0.05.

\*\* 0.001 < p < 0.01.

\*\*\* p < 0.001.

As it can be seen from Table 8, the variables that concurred in identifying GPs with a higher knowledge about the SIDS risk factors can be divided into professional and personal. From a professional perspective, the absence of the variable identifying those GPs holding a post-graduate title in child health and paediatrics is surprising. The syllabuses of the programmes for titles such as the Diploma in Child Health, in fact, specifically include SIDS and its risk factors. Instead, what significantly increased GPs knowledge was direct experience of a case of SIDS, while age, which was highly correlated with seniority (Spearman's  $\rho=0.894$ ,  $p<0.001$ ), tended to have a detrimental effect.

Considering other personal characteristics, it was possible to see how the factors that could influence the personal interest of GPs on this topic (such as being a woman and having children) played a major role. The highest differences between the percentiles were recorded for the variables about having children and being female, which may represent some good proxy variables for the respondents' personal interest in this topic. The median of the index of knowledge in the 'best' situation (female with children, aged 30, with experience about SIDS) was 0.976

(0.934 if normalized between [0,1]). In the 'worst' scenario, instead, (male without children, aged 68, without experience about SIDS) it was -1.319 (0.505 if normalized between [0,1]). Considering the normalized case, the median of the index increased by 85% when moving from the worst to the best profile. The contribution of the explanatory variables was higher at lower percentiles than at the higher ones. In fact, this increase reached 109% at the 20<sup>th</sup> percentile, while it was only 56% at the 80<sup>th</sup>. Having children was always the most important predictor, at every level of the index. The median of the normalized index, with all the other variables being equal to their modal or average value, was 0.748 for practitioners with children and 0.656 for practitioners without children.

### **4.3 GPs' RECOMMENDATIONS TO PARENTS ABOUT INFANT SLEEP POSITIONING**

In the last step of our analysis we investigated the recommendations of GPs regarding infant sleep positioning.

Not all GPs stated that they discussed the SIDS issue with parents (211, 60.5%). In order to properly consider this selection process, we modelled our data with a sample selection model for binary data.

When we analysed the profile of the GPs discussing this issue with parents, it emerged that having directly experienced a case of SIDS was an important determinant (the coefficient 0.43 - Table 9 - implies a risk ratio of 1.30). Moreover, it was very interesting to observe the regional effect that emerged associated with the county of Hampshire (excluding the cities of Portsmouth and Southampton – grouped with the county of the Isle of Wight). From Table 9, we can see that GPs in Hampshire were more likely to discuss this issue with parents (with a risk ratio of 1.38 with respect to Berkshire). Indeed, 70.2% of GPs from Hampshire talked with parents, versus 55.7% of GPs from other counties. However, the SIDS rate in Hampshire (0.20 in 2011 [data retrieved from the Vital Statistics Tables produced by the Office for National Statistics]) is not the highest in the region (it was 0.37 in Berkshire) and it is not higher than the average country level for England and Wales (0.40 in 2010). As a consequence, this result cannot be explained by more attention given to the problem due to a high incidence of SIDS. Instead, it might be explained by the fact that in many areas of Hampshire a prevention campaign named Safer Babies has been carried out since 2008 (when the 'SIDS 10<sup>th</sup> International Conference' was held in Portsmouth, Hampshire) in a bid to reduce the risk of SIDS.

**Table 9: Heckman's sample selection model: estimates of GPs' frequency in talking with parents about the safest position for infants and their correct recommendation**

Selection model: GP talks at least once a month with parents about the correct sleep position			
Variable	Coefficient	Std. Err.	Significance
Intercept	-0.307	0.192	0.110
Gender (female)	0.268	0.140	0.056
Direct experience of a case of SIDS	0.427	0.153	<b>0.005</b>
County (Ref: Berkshire)			
Buckinghamshire	0.274	0.305	0.370
Hampshire	0.507	0.223	<b>0.023</b>
Oxfordshire	0.022	0.231	0.925
Isle of Wight <sup>a</sup>	0.261	0.245	0.286
Main model: GP gives the correct recommendation about the infant's sleep position			
Variable	Coefficient	Std. Err.	Significance
Intercept	2.370	0.940	<b>0.012</b>
Gender (female)	0.808	0.354	<b>0.023</b>
GP works in only one practice	1.387	0.357	<b>0.000</b>
Average number of colleagues	-0.130	0.059	<b>0.029</b>
Rho <sup>b</sup>	0.098	0.984	0.921

<sup>a</sup> The cities of Portsmouth and Southampton were included in the Isle of Wight County to balance the county sample sizes.

<sup>b</sup> Rho is the correlation between the error terms of the two models

Women were more likely to give parents a recommendation about the babies' sleep position and to recommend exclusively the supine position. As it was observed in the first step of our analysis, GPs who worked in only one practice gave a more correct recommendation than those who worked in two or more (with a coefficient of 1.39, corresponding to a risk ratio of 1.37). The importance given to the variable expressing the average number of colleagues might go in the same direction: a higher number of colleagues could imply a lower regular presence of a specific GP in the practice, meaning that this GP might have a lower chance of being chosen as 'reference GP' by parents with children.



## 5. DISCUSSION

This research presents for the first time an analysis of GPs' awareness of the importance of babies' sleep position and other risk factors for SIDS in the United Kingdom. Additionally, it tries to identify those GPs who exclusively recommended the supine position and those who did not.

Over the last 20 years many prevention campaigns have been carried out to spread awareness that the supine position is the safest position for preventing SIDS. Still, approximately 13% of GPs did not give the correct answer about the safest sleep position (the most fundamental aspect in this topic), meaning that there is still room for further increasing awareness.

We showed that GPs with lower seniority tended to have a better knowledge about the safest sleep position, suggesting that their older colleagues may not have been recently updated on the latest scientific evidence.

Moreover, those GPs working in a single practice also presented a higher likelihood of having correct knowledge. We believe this might be the result of mothers preferring seeking more the advice of the GP they have always dealt with rather than that of the GPs who are only seldom at the surgery.

In terms of overall knowledge about SIDS risk factors, we were surprised not to see the variable identifying those GPs holding a post-graduate title in child health and paediatrics among the significant explanatory variables. Instead, direct experience of a case of SIDS significantly increased the GPs' knowledge, while seniority had a detrimental effect once again. This could suggest that once GPs have received their initial training on SIDS and its risk factors they do not get any further follow-up, thus being at risk of not receiving the latest updates.

'Being a woman' and 'Having children' strongly and positively contributed to knowledge. Moreover, when we looked at the recommendations given to parents by GPs, we noticed that those GPs who had direct experience of a case of SIDS were more likely to discuss this issue with parents. All these results seem to confirm the importance of physicians' direct interest in this topic.

We also noticed a regional effect for the county of Hampshire, which might be the result of the prevention campaign named Safer Babies, carried out since 2008 in many areas of the county. This would be very encouraging in terms of evaluation of the effectiveness of prevention campaigns, and it is in line with similar regional effects that were found in Italy (de Luca and Boccuzzo, 2014).

Among those GPs who discussed the correct babies' sleep position with parents, those who were more likely to recommend exclusively the supine position were women and those who worked in only one practice and had a limited number of colleagues.

In conclusion, we think that policy actions are necessary in order to reduce the rate of GPs with knowledge about the sleep position which is not in line with official

guidelines. The effectiveness of previous campaigns is a stimulus in this direction. At present, personal characteristics and interest are the strongest determinants for a correct knowledge on this topic. This could be seen as a challenge to policy makers, as these personal variables represent factors on which policy makers have limited influence. Conversely, variables on which they could have a greater influence proved less effective in determining respondents' knowledge on the topic (e.g., obtaining a post-graduate title in child health and paediatrics). This could imply that there is a need to review the post-graduate trainings' modules about SIDS and the measures in place for ensuring the GPs are informed with the latest scientific evidence.

Further contributions are much needed, especially regarding the knowledge of midwives and health visitors on this topic, as they are two fundamental professional figures who are also in charge of transmitting the SIDS prevention message to parents. It would also be very interesting to see whether the conclusions that were drawn in this article apply to the other regions of the country.

This study has also some limitations: the response rate was quite low, although it was higher than in most of the studies on this topic that were found in the literature. The use of a 'token of appreciation' could have helped in increasing the response rate, but its use was not possible due to budget constraints. Considering the increasingly high use of the Internet that also involves healthcare professionals, we highly recommend, if possible, having the list of respondents' email addresses. In this way, a third reminder would be possible and some respondents might prefer to respond by email rather than using pen and paper. Another limitation might be given by the fact that the sample frame retrieved from the website of the National Health Service (NHS) had been updated about 17 months before the survey started. This could have been the source of some bias, especially in terms of retired doctors and newly employed ones. Unfortunately, it was not possible to take any action in order to prevent or reduce this potential bias.

## ACKNOWLEDGMENTS

This study is part of the SIDS Project, a project of the University of Southampton that explores the dissemination of knowledge about SIDS and its risk factors among healthcare professionals in Spain, Italy and the United Kingdom. The survey was carried out thanks to the contributions made by the Southampton University Strategic Research Development Fund and the Parkes Foundation. We gratefully acknowledge the contribution of Mercedes Burgos Andrés, Omar De La Riva, Rossella Icardi and Marcos Gomez Mella to the mailing operations of this survey.

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