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Five-Year Follow-up of Harms and Benefits of Behavioral Infant Sleep Intervention: Randomized Trial

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KEY WORDS

mental health, attachment, stress, psychological, sleep disorders, population surveillance, child, preschool, cluster randomized controlled trial

ABBREVIATIONS

aOR—adjusted odds ratio

SDQ—Strengths and Difficulties Questionnaire

Drs Wake and Hiscock conceived the original study; Dr Price led the 6-year-old data collection phase; Drs Price, Wake, Ukoumunne, and Hiscock wrote the manuscript; all authors had full access to all of the data (including statistical reports and tables) in the study and take responsibility for the integrity of the data and the accuracy of the data analysis; Dr Price is the guarantor.

This trial has been registered with the ISRCTN Register (<http://isrctn.org>) (identifier ISRCTN48752250).

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WHAT'S KNOWN ON THIS SUBJECT: Behavioral techniques effectively reduce infant sleep problems and associated maternal depression in the short- to medium-term (4–16 months' postintervention). Despite their effectiveness, theoretical concerns persist about long-term harm on children's emotional development, stress regulation, mental health, and the child-parent relationship.



WHAT THIS STUDY ADDS: Behavioral sleep techniques did not cause long-lasting harms or benefits to child, child-parent, or maternal outcomes. Parents and health professionals can feel comfortable about using these techniques to reduce the population burden of infant sleep problems and maternal depression.

abstract

BACKGROUND AND OBJECTIVES: Randomized trials have demonstrated the short- to medium-term effectiveness of behavioral infant sleep interventions. However, concerns persist that they may harm children's emotional development and subsequent mental health. This study aimed to determine long-term harms and/or benefits of an infant behavioral sleep program at age 6 years on (1) child, (2) child-parent, and (3) maternal outcomes.

METHODS: Three hundred twenty-six children (173 intervention) with parent-reported sleep problems at age 7 months were selected from a population sample of 692 infants recruited from well-child centers. The study was a 5-year follow-up of a population-based cluster-randomized trial. Allocation was concealed and researchers (but not parents) were blinded to group allocation. Behavioral techniques were delivered over 1 to 3 individual nurse consultations at infant age 8 to 10 months, versus usual care. The main outcomes measured were (1) child mental health, sleep, psychosocial functioning, stress regulation; (2) child-parent relationship; and (3) maternal mental health and parenting styles.

RESULTS: Two hundred twenty-five families (69%) participated. There was no evidence of differences between intervention and control families for any outcome, including (1) children's emotional ($P = .8$) and conduct behavior scores ($P = .6$), sleep problems (9% vs 7%, $P = .2$), sleep habits score ($P = .4$), parent- ($P = .7$) and child-reported ($P = .8$) psychosocial functioning, chronic stress (29% vs 22%, $P = .4$); (2) child-parent closeness ($P = .1$) and conflict ($P = .4$), global relationship ($P = .9$), disinhibited attachment ($P = .3$); and (3) parent depression, anxiety, and stress scores ($P = .9$) or authoritative parenting (63% vs 59%, $P = .5$).

CONCLUSIONS: Behavioral sleep techniques have no marked long-lasting effects (positive or negative). Parents and health professionals can confidently use these techniques to reduce the short- to medium-term burden of infant sleep problems and maternal depression. *Pediatrics* 2012;130:643–651

Infant sleep problems are prevalent, reported by up to 45% of mothers in the second 6 months of life, and double the risk of maternal depression symptoms.^{1,2} As a common driver of health care use during infancy, they are also costly for families and health systems.³ Fortunately, they are often readily treatable. From 6 months of age, extinction-derived behavioral techniques like “controlled comforting” (see D1 in Guide for Fig 1) help infants learn to self-settle and sleep independently.^{4,5}

Mindell et al's 2006 systematic review of behavioral interventions for child sleep problems found that 49 of 52 programs led to clinically significant reductions in bedtime resistance and night waking 3 to 6 months later.⁴






























Secondary benefits included better parent sleep, mental health, and child-parent relationships. No studies, including the longest follow-up to date (3 years' postintervention),⁶ have reported detrimental effects.^{3,4,7,8} The American Academy of Sleep Medicine subsequently classified behavioral techniques as “standard” practice for managing infant sleep problems,⁹ and a recent expert working party convened by the Australian Research Alliance for Children and Youth concluded that behavioral approaches are safe, at least in the short term.¹⁰

Despite strong evidence of the short- and medium-term benefit and cost-effectiveness of behavioral sleep techniques, unproved concerns about

their long-term harm are limiting their uptake and provoking vigorous debate. For example, a 2011 review by Blunden et al¹¹ notes that behavioral techniques could prevent parents from responding consistently and sensitively to their child, thereby leading to long-term adverse impacts on child-parent bonding, child stress regulation, mental health, and emotional development. These concerns originated with pure extinction (“crying-it-out”), which is not usually recommended nowadays because of the distress it causes parents and infants. However, the concerns have extended to extinction derivatives like controlled comforting and “camping out,”¹¹ which are recommended for best practice.^{4,9} In response to Blunden et al,¹¹ Sadeh et al¹² countered that there is no evidence that behavioral techniques cause harm. Researchers from these opposing perspectives are calling for a rigorous longitudinal study of the long-term effects of behavioral sleep interventions to resolve this controversy.^{11,12}

Interestingly, this debate is largely framed around possible harms rather than the potential for lasting benefits. In the absence of long-term follow-up studies, it is entirely possible that benefits to maternal mental health may extend beyond the medium-term already demonstrated.⁷ Furthermore, teaching parents to regulate their children's sleep behavior is a form of limit setting that, combined with parental warmth, constitutes the optimal, authoritative, parenting style for child outcomes.^{13,14}

In 2003–2005, Hiscock and colleagues³ conducted the Infant Sleep Study. Designed to improve Australian infants' sleep problems at 8 to 10 months of age, it was a large, community-based, secondary-prevention randomized trial of a behavioral intervention comprising positive bedtime routines and teaching either controlled comforting

Date	Age of child	Intervention group	Control group
Oct to Nov 2003	4 months	 	 
Jan to Feb 2004	7 months		
		Randomization	
			
Feb to Mar 2004	8 months	 	
	8–10 months		
Apr to May 2004	10 months		
Jun to Jul 2004	12 months	 	 
Dec 2004 to Jan 2004	18 months		
Jun to Jul 2005	24 months	 	 
Dec 2007 to Jan 2006	3.5 years		
Jun to Jul 2009	6 years	 	 

Guide for Figure 1







	Scheduled, universally available, state-funded maternal and child health (MCH) nurse visit to offer support, information and advice relevant to child's developmental stage. At 4 months, this included an invitation to express interest in the study, followed by researcher contact and trial recruitment if interested.
	Maternal written questionnaire
	Intervention MCH nurses attended two 2.5 hour training sessions to learn how to administer the intervention.
	Intervention mothers reporting an infant sleep problem consulted with their MCH nurse at the usual 8-month visit. They discussed normal sleep patterns, the nature of sleep problems and potential causes, identified potential solutions and developed an individualized sleep management plan. Mothers began completing a 2-week sleep diary. Two main sleep techniques were offered: (1) “controlled comforting,” in which parents respond to their infant's cry at increasing time intervals to allow the child to learn to self-settle, and (2) “camping out,” in which the parent sits with the child as they learn to independently fall asleep, slowly removing their presence from the child's room.
	Mothers attended up to 2 follow-up MCH nurse consultations to help recognize sleep patterns and improvements, and set further goals.
	Home visit conducted to administer the child-reported Pediatric Quality of Life Inventory and practice collection of salivary cortisol.

FIGURE 1
Graphical depiction (“Perera diagram”³⁰) of the components of the trial shared and unique to the intervention and control groups. MCH, maternal and child health.

or adult fading (also known as camping out), should parents choose to use them. In comparison with controls, intervention parents reported fewer sleep problems at infant age 10 months (56% [intervention] vs 68% [control]; adjusted odds ratio [aOR] 0.6 [95% confidence interval 0.4–0.9]) and 12 months (39% vs 55%; aOR 0.5 [0.3–0.8]),³ with a sustained reduction in maternal depression at 2 years (15% vs 26%; aOR 0.4 [0.2–0.9]).⁷

To determine long-term harms and/or benefits of this infant behavioral sleep intervention, we now report our 2009 follow-up at age 6 years. We hypothesized that there would be no evidence of intervention versus control group differences in: (1) child emotional and conduct behavior (primary outcomes), sleep, psychosocial health-related quality of life, and diurnal cortisol as a marker of stress; (2) child-parent relationship, disinhibited attachment; or (3) maternal mental health or parenting styles.

METHODS

Design and Setting

The Kids Sleep Study is the 5-year follow-up of the Infant Sleep Study, a randomized controlled trial (International Standard Randomized Controlled Trial Number 48752250) for which we have previously reported methods for outcomes at ages 12³ and 24⁷ months. In brief, the Infant Sleep Study aimed to recruit all mothers with children born in June to July 2003 who attended the free, universal 4-month well-child check with their maternal and child health nurse in 6 sociodemographically diverse local government areas ($n = 982$). Of these, 782 (80%) expressed interest in participating; 692 (70%) returned the 7-month questionnaire. Mothers who responded “Yes” to the 7-month screening questionnaire item “Over the last 2 weeks, has your baby’s sleep generally been a problem for

you?” were eligible for the trial ($n = 328$, 47%). Maternal and child health nurses excluded infants born <32 weeks’ gestation and mothers with insufficient English to complete questionnaires.

After baseline recruitment, we randomized the 49 maternal and child health nurse centers (clusters) which, in turn, determined participant allocation.³ Because nurses were responsible for delivering the intervention, randomizing clusters rather than participating families minimized the likelihood of contamination between trial arms. Centers were ranked within each stratum according to the number of infants recruited at 4 months, randomizing the largest center and alternately allocating subsequent ones to avoid a marked imbalance in cluster sizes between trial arms. Because all the centers were recruited before randomization and ranked by using a criterion that could not be influenced by the investigators, allocation concealment was achieved. Researchers involved in data collection and entry were blinded; nurses and parents, however, could not be blinded to group allocation.

Intervention nurses were trained to deliver a brief, standardized behavioral sleep intervention at the routine 8-month well-child check to mothers reporting infant sleep problems (Fig 1 and Guide show details). Based on their needs and preferences, each family chose which (if any) type or mix of strategies they would use to try and manage their infant’s sleep.³ One hundred of the 174 intervention mothers attended their nurse well-child check visits to discuss infant sleep problem management for an average of 1.52 visits, with mean duration for the first and subsequent visits of 25 and 19 minutes, respectively. Control families received usual care, which meant they were free to attend the scheduled 8-month visit and ask for sleep advice; control nurses, however, were not

trained to deliver specific sleep management techniques.

Follow-up Patients and Procedures

From April to October 2009, we recontacted all families. Of the original 328 Infant Sleep Study children,³ 326 were eligible at age 6, whereas 2 met our prespecified exclusion criteria of intellectual disability or developmental delay (Fig 2). Parents who returned written informed consent were mailed a questionnaire and phoned to arrange a 40- to 60-minute home-based assessment as close as practicable to the child’s sixth birthday, during which the trained researchers (1) administered the Pediatric Quality of Life Inventory¹⁵ to the child and (2) showed families how to collect salivary cortisol (see Table 1).

Families selected a nonschool day (weekend or holiday) to collect 2 cortisol samples: (1) 30 to 40 minutes after waking to avoid the postawakening rise, because its meaning in relation to the diurnal cortisol profile or psychosocial stress is unclear,¹⁶ and (2) before lunch. We based our collection protocol on the standardized procedures provided by the pathology laboratories responsible for testing samples. Children avoided brushing teeth, eating or drinking for 30 minutes before collection, then thoroughly rinsed their mouth with water 3 times, chewed a piece of Wrigley’s sugarfree gum, EXTRA peppermint, and collected 4 mL of saliva in a plain tube. Families recorded children’s waking and saliva collection times. Families stored samples at room temperature before mailing them back within 1 to 2 weeks of collection,¹⁷ when we froze them at -18°C . Cortisol levels were measured by 2 local laboratories owing to an unexpected company merger (by using the Roche Modular and Avida Centaur systems, respectively). Interassay coefficients of variation fell below 5.3%

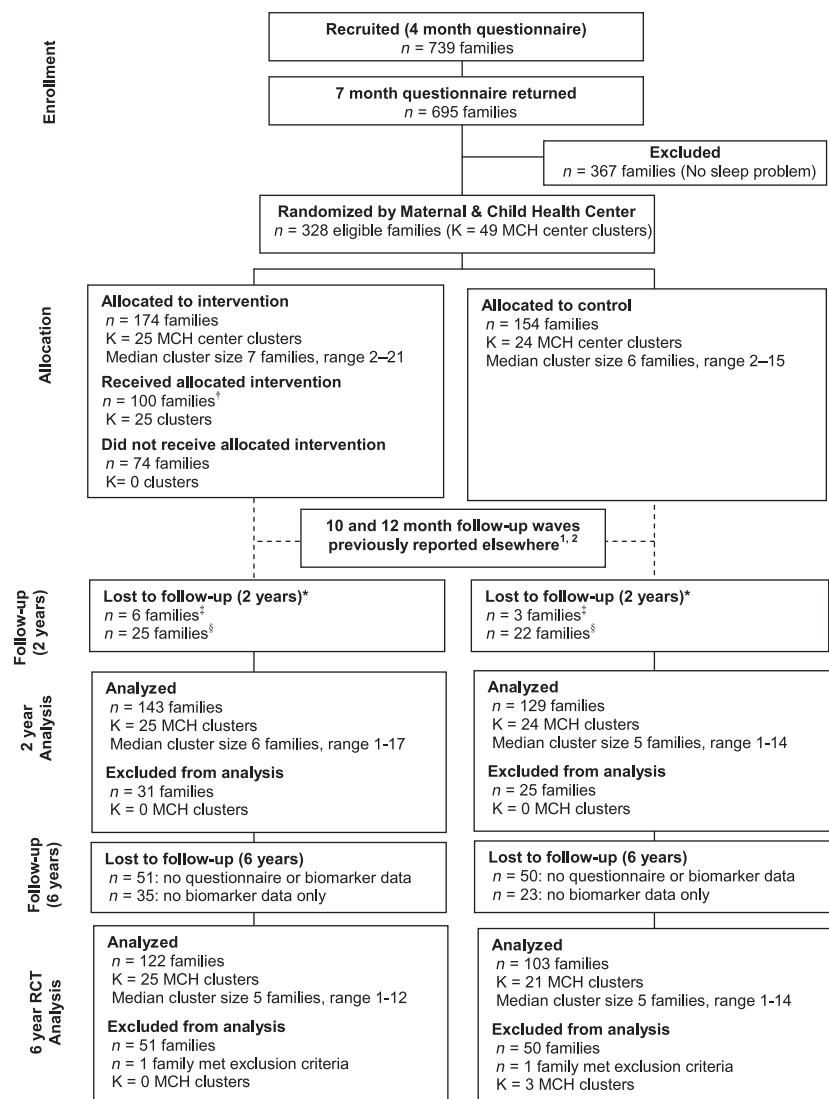


FIGURE 2

Participant flow for the original Infant Sleep Study to 6-year-old outcomes. All clusters were trained (so 0 “did not receive intervention”), but not all individuals received the intervention. [†]Take-up of the intervention was voluntary. One hundred families reported receiving the intervention. ^{*}All lost to follow-up because of failure to return questionnaires. [‡]Did not return the 10- or 12-month follow-up questionnaire (were not sent 2-year questionnaire). [§]Did not return 2-year follow-up questionnaire. MCH, maternal and child health.

($n = 113$ samples) and 15% ($n = 54$) for the 2 laboratories, respectively. No saliva-based intraassay reliabilities were available. The proportion of intervention samples analyzed by each laboratory was similar (55% vs 46%, respectively).

Measures

Table 1 shows details of the outcome measures. For all variables but cortisol, we selected potential confounding

variables a priori based on existing research.¹⁸ Throughout childhood, child gender, temperament, maternal depression, and socioeconomic status (maternal education and Socioeconomic Indexes for Areas¹⁹ Index of Relative Disadvantage) are associated with and predict most outcomes examined in the Kids Sleep Study.^{1,20} Socio-Economic Indexes for Areas is a national index derived from census data for all individuals living in

a postal code, with higher scores indicating less disadvantage. We controlled for all 5 potential confounders in the adjusted analyses, except some analyses of binary outcomes in which the sample size was too small to include all 5 without causing instability in the resulting estimates²¹ (see “Sample Size and Analyses” below). For Strengths and Difficulties Questionnaire (SDQ) continuous scores, we additionally adjusted for financial stress (6-point quantitative item, “Given your current needs and financial responsibilities, how would you say you and your family are getting on?”; responses range from “prosperous” to “very poor”), because Longitudinal Study of Australian Children data indicate that financial stresses are associated with a doubling of the risk of behavior problems in 2- to 7-year-olds.²² In the absence of a strong conceptual framework for choosing cortisol confounders, we used exploratory analyses to identify variables associated with “abnormal” cortisol levels at $P < .1$.¹⁸ This applied to the laboratory at which testing occurred only.

Sample Size and Analyses

The original Infant Sleep Study was powered to detect a difference of 20% between the proportions of mothers reporting infant sleep problems at each of the 10- and 12-month follow-ups with 80% power at the 5% level of significance, with an assumed cluster size of 11 and intracluster correlation coefficient of 0.02.³ For the Kids Sleep Study follow-up (not considered at the original sample size calculation), we anticipated retaining at least 75% of the 2-year-old participants (99 of 132 control and 110 of 146 intervention families, total $n = 209$). A sample size of 99 per group would give the study 80% power to detect a difference of 0.4 SD units (ie, effect size) between groups at the 5% level of significance. We did not allow for intracluster correlation, because we

TABLE 1 6-Year-Old Outcome Measures; Parent Report Unless Otherwise Specified

Construct	Measure
Child	
Emotional and behavior problems (primary outcomes)	25-item SDQ (SDQ 4- to 10-y-old version). ³⁵⁻³⁶ Emotional and conduct behavior, and total difficulties scores analyzed as continuous (higher scores indicate greater problem) and binary: dichotomized into clinical (top 10%) versus normal (bottom 90%) according to Australian normative data for 6-y-old boys and girls (cutoff scores as follows: total: ≥ 16 and ≥ 14 ; emotional: ≥ 5 and ≥ 4 ; conduct behavior: ≥ 4 and ≥ 3 , respectively). ²⁷
Perception of sleep as a problem	Single item from the Longitudinal Study of Australian Children. ³⁶ Presence of a sleep problem (no, mild, moderate, or severe problem) dichotomized into “no/mild” versus “moderate/severe” problem. ³⁵
Clinical sleep problem	33-item Child Sleep Habits Questionnaire Short Form (CSHQ). ³⁰ Scale analyzed as continuous; higher scores indicate greater problem. ³⁰
Psychosocial health-related quality of life (HRQoL)	23-item Pediatric Quality of Life Inventory 4.0 (PedsQL 4.0), ³⁶ parent-proxy and self report (collected by interview) 5- to 7-y-old versions. ¹⁵⁻³¹ Higher scores represent better HRQoL.
Stress	Implicit in the theoretical concerns that behavioral sleep techniques impair children's subsequent emotional development is the notion that they subject the child to extreme, irresolvable stress. ¹¹ We chose to collect cortisol as an objective measure of children's stress because collection is simple and noninvasive. ³⁷ The normal cortisol profile is highest upon waking, shows a steep descent over the morning, and then a shallower descent to the nadir at around midnight. We collected waking and lunchtime salivary cortisol samples to capture 2 abnormal profiles evident in samples of children exposed to exceptional early stress: (1) “hypo” (starts low and continues low) and (2) “hyper” (starts high and continues high). ³⁸⁻³⁹ Because there is no consensus on how to quantify these profiles, we divided children's waking cortisol and waking-to-lunch hourly change score (Δ WL/h) into internal tertiles, categorizing children with low waking cortisol and low Δ WL/h (ie, “hypo”; $n = 34$), and high waking cortisol and low Δ WL/h (ie, “hyper”; $n = 4$) as “abnormal,” for comparison against the remaining “normal” sample ($n = 111$). Excluding the 4 “hyper” children did not alter the results, so they were retained in the analyses.
Child-parent	
Perception of child-parent relationship	15-item Child-Parent Relationship Scale Short-Form (CPRS) ²⁹ Closeness and conflict subscales; higher scores indicate greater closeness or conflict.
Global rating	Study-designed 5-point item (“How would you rate your current relationship with this child?”). Higher scores indicate better relationship.
Disinhibited attachment	From 5-item disinhibited attachment interview, ⁴⁰ scored on the CPRS scale. Higher scores indicate increased disinhibited attachment.
Maternal	
Depression, anxiety, stress	21-item Depression, Anxiety, Stress Scale Short-Form (DASS). ²⁸ Subscale scores are doubled and transformed as specified on the DASS Web site (http://www2.psy.unsw.edu.au/groups/dass/) to approximate the complete scale. The total score sums the 3 subscale scores. Higher total and subscale scores indicate worse mental health.
Parenting styles	11 items from the Longitudinal Study of Australian Children ¹⁴ on 2 subscales, Warmth ⁴¹ and Control, ⁴² which combine to measure 4 key parenting styles: (1) authoritative (high warmth, high control: optimal parenting ¹⁴); (2) authoritarian (low warmth, high control); (3) permissive (high warmth, low control); and (4) disengaged parenting (low warmth, low control). ¹³

expected any cluster effects to fade over the 5 years since the intervention.

We compared trial arms by fitting random effects linear regression models estimated by using maximum likelihood for quantitative outcomes, and marginal logistic regression models by using generalized estimating equations, assuming an exchangeable correlation structure with information sandwich (“robust”) estimates of SE for binary outcomes.²³ Both methods allow for correlation between outcomes of participants from the same cluster. We conducted analyses unadjusted and adjusted for the potential confounders, with the exception of analyses of (1) SDQ binary outcomes, which was not

adjusted for maternal education, and (2) child “moderate/severe” sleep problem, which was not adjusted for child gender, maternal depression, or education, because there were potentially too few subjects with clinically high SDQ scores or a sleep problem to obtain stable estimates from models with all potential confounders included as predictors. The omitted variables were not strongly related to the respective binary outcomes.

All retained participants were analyzed in the groups to which they were randomized, applying the intention-to-treat principle. Confidence intervals from analyses of quantitative outcomes were validated by using the bootstrap

method.²⁴ Intraclass (intra-maternal and child health unit) correlation coefficients from adjusted analyses are reported according to the CONSORT recommendations for cluster-randomized trials.^{25,26} All data files were analyzed by using Intercooled Stata, version 11.1 for Windows (Stata Corp, College Station, TX).

Both the original trial (23067B) and 6-year-old follow-up (28137F) were approved by the Human Research Ethics Committee of The Royal Children's Hospital, Melbourne.

RESULTS

At age 6 years, 225 of 326 children (69%) participated (see Fig 2, participant

flow). Of these, 193 (86%) participated in the home visit and 177 (79%) agreed to collect cortisol. Of the latter, 167 (94%) provided at least 1 cortisol sample and 149 (84%) provided the 2 cortisol samples and the collection time data required to categorize the diurnal profile as “abnormal” versus “normal.” We were unable to contact 49 of 326 families (15%), and 52 of 326 (16%) families declined for reasons including “too busy” ($n = 26$), “not interested” ($n = 6$), “personal reasons” ($n = 6$), “child illness” ($n = 1$), or no reason ($n = 13$).

Table 2 shows the sample characteristics. In the control arm, children of mothers who completed a university degree were overrepresented, and children from disadvantaged backgrounds were underrepresented among those retained versus lost to follow-up; children of families who spoke a language other than English at home were underrepresented in both arms. Follow-up occurred at a mean age of 6.0 years (SD 1.9 months). Of the retained families, those who did and did not collect at least 1 cortisol sample had similar baseline characteristics (data available

from authors on request), with the exception that those who did were less likely to speak a language other than English at home (13% vs 26%).

There was little evidence of unadjusted or adjusted differences between trial arms on the child, child-parent, and maternal outcomes (Table 3). Mean scores were almost identical between groups for the parent-reported child emotional, conduct behavior, and total mental health difficulties; Child Sleep Habits Questionnaire; psychosocial health-related quality of life; the child-parent relationship measures; and maternal mental health. The proportions of children with mental health problems, “moderate/severe” sleep problems, and authoritative parenting were also similar between trial arms. Consistent with these findings, the mean scores for children’s self-reported health-related quality of life and the proportions of children classified with chronic stress according to the objective physiologic cortisol measure were similar between intervention and control groups, providing little evidence that the early intervention harmed or benefited the

intervention group with respect to child, child-parent, or maternal outcomes at 6 years.

DISCUSSION

There was no evidence that a population-based targeted intervention that effectively reduced parent-reported sleep problems and maternal depression during infancy had long-lasting harmful or beneficial effects on child, child-parent, or maternal outcomes by 6 years of age. Thus, this trial indicates that behavioral techniques are safe to use in the long-term to at least 5 years postintervention.

The study had several strengths. This 5-year follow-up of a rigorously conducted randomized trial (the gold standard for assessing causality)¹⁸ may represent the only opportunity to provide objective evidence investigating any lasting harms or benefits of behavioral infant sleep interventions. This is because, with their known short- and medium-term effectiveness, it is unlikely that new trials with true non-intervention controls and 5-year follow-up could now be ethically

TABLE 2 Baseline Characteristics According to Follow-up Status (ie, Retained or Lost to Kids Sleep Study) at Age 6 Years

Baseline Characteristics (4 mo)	Total (N = 326)		Intervention (n = 173)		Control (n = 153)	
	Retained (n = 225)	Lost (n = 101)	Retained (n = 122)	Lost (n = 51)	Retained (n = 103)	Lost (n = 50)
Child						
Male	54.7	53.5	50.8	52.9	59.2	54.0
Age (months), mean (SD)	4.6 (0.6)	4.6 (0.6)	4.6 (0.5)	4.7 (0.6)	4.5 (0.7)	4.5 (0.7)
Difficult temperament	24.4	27.7	23.0	21.6	26.2	34.0
Mother						
Age (years), mean (SD)	33.1 (4.4)	32.9 (4.9)	32.6 (4.2)	33.4 (4.5)	33.6 (4.6)	32.3 (5.1)
Depression (EPDS), mean (SD)	8.5 (5.1)	8.2 (5.3)	8.4 (5.1)	8.3 (5.6)	8.5 (5.2)	8.2 (5.0)
Depression (EPDS) >9	39.6	39.6	36.1	41.2	43.7	38.0
Education status						
Did not complete high school	15.6	21.8	15.6	19.6	15.5	24.0
Completed high school	30.2	35.6	30.3	31.4	30.1	40.0
University degree	54.2	42.6	54.1	49.0	54.4	36.0
Family						
Index of Social Disadvantage						
High disadvantage	15.1	24.8	13.9	13.7	16.5	36.0
Medium disadvantage	33.3	26.7	36.9	33.3	29.1	20.0
Low disadvantage	51.6	48.5	49.2	52.9	54.4	44.0
Language other than English	16.0	26.0	14.8	25.5	17.5	26.5

All values are percentages, except where otherwise stated. EPDS, Edinburgh Postnatal Depression Scale, where EPDS >9 is the community cut point for depression; SEIFA, Socioeconomic Indexes for Areas, 2002 Australian census data for socioeconomic status by postal code.

TABLE 3 Results of Regression Analyses Comparing the 2 Trial Arms on Child, Child-Parent, and Maternal Outcomes at Age 6 Years

Outcome	Descriptive Statistics ^a				Comparative Statistic: I Compared With C ^b (95% CI)				
	Intervention (I)		Control (C)		Unadjusted Statistic	Adjusted			ICC ^c
	<i>n</i>	Summary	<i>n</i>	Summary		Statistic	95% CI	<i>P</i>	
Child									
SDQ Total score	122	8.5 (5.7)	103	8.1 (6.0)	0.4	0.5	−1.0 to 1.9	.5	0
SDQ Total problems, %	122	11.5	103	16.5	0.7	0.6	0.3 to 1.2	.2	0
SDQ Emotion score	122	1.8 (2.0)	103	1.8 (2.0)	0.03	−0.04	−0.6 to 0.5	.8	0
SDQ Emotion problems, %	122	12.3	103	16.5	0.7	0.5	0.3 to 1.0	.07	0
SDQ Conduct behavior score	122	1.8 (1.8)	103	1.8 (1.8)	0.1	0.1	−0.3 to 0.6	.6	0
SDQ Conduct behavior problems, %	122	22.1	103	23.3	0.9	0.9	0.5 to 1.8	.8	0.0003
Sleep problem, %	122	9.0	102	6.9	1.6	1.6	0.8 to 3.1	.2	0
CSHQ Total	115	42.2 (6.1)	97	42.7 (8.1)	−0.5	−0.8	−2.7 to 1.1	.4	0
PedsQL Psychosocial, parent-proxy	122	78.9 (12.0)	103	78.3 (13.9)	0.7	0.5	−2.7 to 3.8	.7	0.006
PedsQL Psychosocial, child-self	98	69.1 (15.3)	88	68.6 (16.8)	0.4	0.6	−3.8 to 5.0	.8	0
“Abnormal” cortisol, %	80	28.8	69	21.7	1.4	1.4	0.6 to 3.3	.4	0.02
Child-parent									
CPRS Closeness	122	4.3 (0.3)	103	4.3 (0.2)	−0.1	−0.04	−0.1 to 0.01	.1	0
CPRS Conflict	122	2.3 (0.8)	103	2.2 (0.8)	0.1	0.1	−0.1 to 0.3	.4	0
Global rating	121	4.5 (0.6)	103	4.5 (0.7)	−0.03	−0.01	−0.2 to 0.2	.9	0
Disinhibited attachment	122	2.9 (0.9)	103	3.1 (1.0)	−0.2	−0.1	−0.4 to 0.1	.3	0
Maternal									
DASS Total score	122	17.3 (11.8)	103	17.5 (16.4)	−0.1	−0.4	−3.7 to 2.9	.9	0
DASS Depression	122	4.8 (4.9)	103	4.7 (5.9)	0.1	−0.1	−1.6 to 1.4	.9	0.06
DASS Anxiety	122	2.8 (3.6)	103	3.4 (5.6)	−0.5	−0.6	−1.7 to 0.6	.4	0.003
DASS Stress	122	9.7 (5.9)	103	9.4 (6.7)	0.3	0.3	−1.2 to 1.8	.7	0
Authoritarian/permissive/disengaged parenting style, ^d %	122	36.9	103	40.8	0.9	0.8	0.5 to 1.4	.5	0.004

CI, confidence interval; CPRS, Child-Parent Relationship Scale; CSHQ, Child Sleep Habits Questionnaire; DASS, Depression, Anxiety, Stress, Scale; ICC, intraclass correlation coefficient; PedsQL, Pediatric Quality of Life Inventory.

^a Summary statistics are quantitative (mean [SD]) except where specified as dichotomous (%).

^b The comparative statistic is the mean difference for quantitative outcomes (intervention minus control) and odds ratio for dichotomous outcomes (the risk of receiving the intervention compared with receiving usual care).

^c Negative ICCs for SDQ Total and Emotional clinical scores, and permissive parenting were truncated at zero.

^d Reference group for parenting styles is authoritative parenting. Authoritarian, permissive, and disengaged parenting styles (all negative outcomes) were similar between groups, and were therefore collapsed into a single category for analysis.

conducted. Where possible, we used well-validated, reliable outcome measures^{15,27–31} collected from multiple sources, including parent report, child report, and objective physiologic biomarkers. Although details of the 30% (290/982) of families originally excluded from the population sample were unknown, the enrolled participants covered a broad socioeconomic range and were similar to Australian and US normative data for maternal well-being and child temperament characteristics,³² meaning that our findings should generalize to English-speaking families.

The study also had some limitations. Because 31% (101/326) of the original sample was lost to follow-up at age 6 years, the lower and upper bounds of

the 95% confidence intervals did not rule out smaller long-term harms or benefits of the intervention that could be meaningful in public health research.⁷ Nonetheless, the precision of the confidence intervals make clinically meaningful group differences unlikely. Loss to follow-up can also introduce internal bias and reduce generalizability. Regarding bias, the retained intervention and control participants were fairly balanced (Table 2); however, as more non-English-speaking and disadvantaged families were lost to follow-up, our findings may be less generalizable to these participant groups. Finally, no validation studies of the categorical cortisol variable were available, but our own exploratory analyses within the combined cohort showing that abnormal cortisol was

associated with poorer child and maternal well-being suggests that it was indeed functioning as a stress biomarker (A.P., M.W., H.H., unpublished data).

Our findings were entirely consistent with the longest follow-up study before the Kids Sleep Study,^{6,20} which reported no differences between intervention and control arms on child internalizing and externalizing problems, sleep, or maternal mental health at child age 3 to 4 years (3 years postintervention). Thus, these new data, when interpreted with shorter follow-up data from >50 intervention studies (including 9 randomized controlled trials), suggest that behavioral sleep interventions have short- to medium-term benefits that fade beyond 2 to 3 years' postintervention.

In the context of potential harm, it is unknown whether there are subgroups of infants (eg, those who have previously been maltreated, experienced early trauma, or are anxious children) for whom the techniques are unsuitable in the short- or long-term.¹² If supported by empirical investigation, there could be a case for using more gradual interventions such as adult fading instead of the more intensive graduated extinction (controlled comforting) to manage infant sleep. Along with trials like ours demonstrating that sleep problems can be effectively treated in older infants, recent efficacy trials for children younger than 6 months suggest that parent education programs that teach parents about normal infant sleep and the use of positive bedtime routines could effectively prevent later sleep problems.^{4,8}

Our findings highlight the importance of access for parents to effective sleep management strategies and training for the health professionals in such

strategies. Currently, the information available to parents about the effects of behavioral sleep strategies is inconsistent and out of date. For example, peak bodies including the Australian Infant Mental Health Association and the Australian Breastfeeding Association, which work to influence policy and practice but argue against the use of behavioral techniques like controlled comforting, have not updated position statements since the mid-2000s. Thus, there is a pressing need to deliver evidence-based information to parents and health care providers, which could be achieved, in part, by updating position statements, policy documents, and training curricula to reflect our current findings that behavioral sleep techniques are both effective in the short- and medium-term and safe to use in the long-term.

CONCLUSIONS

The intervention achieved all of its original aims (better infant sleep and

lower maternal depression and health care costs in the short- to medium-term). The 6-year-old findings indicate that there were no marked long-term (at least to 5 years' postintervention) harms or benefits. We therefore conclude that parents can feel confident using, and health professionals can feel confident offering, behavioral techniques such as controlled comforting and camping out for managing infant sleep.

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Five-Year Follow-up of Harms and Benefits of Behavioral Infant Sleep Intervention: Randomized Trial

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