

SCIENTIFIC INVESTIGATIONS

Behavioral interventions for infant sleep problems: the role of parental cry tolerance and sleep-related cognitions

Michal Kahn, PhD1,*; Efrat Livne-Karp, PhD1,*; Michal Juda-Hanael, PhD1; Haim Omer, PhD1; Liat Tikotzky, PhD2; Thomas F. Anders, MD3; Avi Sadeh, DSc1,†

¹School of Psychological Sciences, Tel Aviv University, Tel-Aviv, Israel; ²Department of Psychology, Ben-Gurion University of the Negev, Beer-Sheva, Israel; ³Brown University, Providence, Rhode Island; *Contributed equally; [†]Deceased

Study Objectives: This study tested whether parental cry tolerance (PCT) and distress-attribution cognitions predict outcomes of behavioral interventions for infant sleep problems. It additionally examined intervention effects on these parental factors.

Methods: Participants were 91 infants aged 9–18 months (61% boys) with sleep-related problems and their parents. Families were randomized to 1 of 2 behavioral interventions for infant sleep problems: Checking-in or Camping-out. Assessments were completed at baseline and 1-month post-treatment. Infant sleep was assessed using actigraphy and parent reports on the Brief Infant Sleep Questionnaire. PCT was measured using the Intervention Delay to Infant Crying Video laboratory paradigm, and parental distress-attribution cognitions were assessed via the Infant Sleep Vignettes Interpretation Scale.

Results: Higher PCT and lower parental distress-attribution cognitions at baseline predicted greater improvement in parent-reported sleep problems post-treatment, and higher PCT additionally predicted larger reductions in the number of reported nighttime awakenings. Moreover, PCT increased, and distress-attribution decreased, following the interventions.

Conclusions: Parent factors both predict and are predicted by behavioral interventions for infant sleep problems. This study's findings suggest that parents with low cry tolerance and high distress-attribution cognitions derive less benefit from these interventions and may thus require augmented care.

Clinical Trial Registration: Registry: ClinicalTrials.gov; Name: Interventions for sleep problems in early childhood; URL: https://clinicaltrials.gov/ct2/show/NCT01489215;Identifier: NCT01489215.

Keywords: infant sleep, behavioral intervention, parenting, cry tolerance, sleep-related cognitions

Citation: Kahn M, Livne-Karp E, Juda-Hanael M, et al. Behavioral interventions for infant sleep problems: the role of parental cry tolerance and sleep-related cognitions. *J Clin Sleep Med*. 2020;16(8):1275–1283.

BRIEF SUMMARY

Current Knowledge/Study Rationale: While evidence for the efficacy of behavioral interventions for infant sleep problems is mounting, many parents are still reluctant to undertake them, find it difficult to adhere to them, or do not benefit sufficiently from them. However, parental factors that may predict intervention outcomes have yet to be examined.

Study Impact: This study showed that parental cry tolerance and sleep-related cognitions predict treatment outcomes, with greater improvement in parent-reported sleep for infants of parents with higher cry tolerance and lower distress-attribution. Furthermore, following treatment, parents demonstrated higher cry tolerance and lower distress-attribution compared with baseline. These findings may inform clinicians in indicating which parent populations may require more intensive support when attempting interventions to improve infant sleep.

INTRODUCTION

Sleep—wake patterns evolve dramatically during infancy. The multiple brief sleep episodes distributed around the clock in most newborn infants usually consolidate into one major nocturnal sleep episode toward 6 months of age. 1.2 However, in 15–30% of infants sleep continues to be fragmented, and sleeplessness persists far beyond the first months of life. 3.4 These infants experience pediatric insomnia, manifested mainly as difficulties initiating and maintaining sleep. 5 Sleep problems such as these have been associated with a multitude of adverse short- and long-term outcomes for children, including physiological, emotional, cognitive, and behavioral problems. 6.7 For parents, consequences include increased risk of maternal depression, poor physical health,

reduced sense of competence, and even thoughts about harming their child.^{8–11}

Fortunately, behavioral interventions for infant sleep problems exist, with established evidence of effectiveness. These techniques stem from psychological learning theory, positing that excessive parental involvement in soothing their infant to sleep hinders the development of infant self-soothing skills, thus perpetuating a dependency on caregivers in commencing and resuming sleep. Parents are encouraged to delay their involvement in soothing the child to sleep as part of these treatments, whether while remaining physically present in the room (as in the Camping-out technique) or withdrawing and returning to check in on the infant at scheduled intervals (as in Graduated Extinction). Findings from the past 3 decades attest to the efficacy of such strategies in improving infant sleep both in the short and

long term. ^{14–16} Furthermore, despite concerns raised by some regarding possible adverse "side effects," no evidence of harm has yet been documented as a result of these interventions. ^{17,18}

Even with mounting evidence for their cost-effectiveness and safety, many parents are still reluctant to undertake behavioral interventions for pediatric insomnia. Of those who do attempt it, 20–30% do not benefit sufficiently. The barriers standing in the way of gaining from such techniques may be manifold. For example, our group recently reported that highly anxious infants were less likely to show improvement in sleep following these interventions, particularly when they included higher degrees of separation from parents. However, parent factors that may hinder implementation of these treatments have yet to be examined.

Parental low ability to tolerate infant distress could be one such barrier, as it has been shown to play a role in the development of pediatric sleep problems. In a cross-sectional study, parental cry tolerance (PCT) was found to be lower in parents of infants with sleep problems compared to parents of infants without sleep problems, and distress-attribution cognitions (ie, the tendency to attribute distress to infants with behavioral sleep problems) were found to be higher in these parents. Moreover, longitudinal studies indicate that lower PCT and higher parental distress-attribution each predict poorer infant sleep. Since behavioral interventions require parents to delay their involvement and tolerate some extent of crying at night, parents who are highly sensitive to infant distress may be less prone to participate or successfully implement such treatments, resulting in poorer outcomes.

These notions dovetail with the growing interest in determining not only which interventions work but also for whom and under which circumstances. In a meta-analysis on treatments for pediatric insomnia, the authors state: "although evidence is strong for behavioral interventions for insomnia in young children, more studies are needed to help identify factors that may predict treatment success". The present study addresses this research gap by examining whether PCT and parental distress-attribution cognitions predict outcomes of behavioral interventions for infant sleep problems.

A further aim of this study was to evaluate the change in PCT and parental distress-attribution following these interventions. Behavioral parent-focused interventions in other child domains have been shown to both be impacted by and have an impact on parents, with positive outcomes including lower parental psychological distress and anxiety and more positive parenting. ^{24–26} More specifically, parent outcomes of behavioral interventions to improve infant sleep have included lower parental depression and more confidence in managing infant sleep. ^{16,27} It is thus likely that these interventions would also increase PCT and decrease distress-attribution cognitions, as a result of psychoeducation, and repeated practice and exposure to child distress within a structured protocol.

In the present study parents of infants with pediatric insomnia were randomly allocated to 1 of 2 interventions: Camping-out or a variation of Graduated Extinction we named Checking-in. Infant sleep, PCT, and parental distress-attribution cognitions were assessed at baseline and post-treatment. The study aimed to test (1) whether PCT and distress-attribution cognitions predict the outcomes of behavioral interventions for infant

sleep problems, (2) whether these interventions affect PCT and distress-attribution cognitions, and (3) whether changes in sleep outcomes are associated with changes in PCT and distress-attribution cognitions. We hypothesized that infants of parents with higher baseline cry tolerance and lower baseline distress-attribution cognitions would show greater improvement in sleep following the interventions. Furthermore, we hypothesized that, compared with baseline, following the intervention parents would be more tolerant to crying and less inclined to interpret impaired sleep as a sign of distress. Finally, we hypothesized that improvement in sleep would be associated with increases in PCT and decreases in distress-attribution cognitions.

METHODS

Participants

The CONSORT (Consolidated Standards of Reporting Trials) diagram in **Figure 1** depicts participant progress through the study stages. Participants were recruited for this study through Web-based media advertisements referring to a study conducted in the pediatric sleep clinic at Tel Aviv University. Inclusion criteria were as follows: (1) infant age range 9–18 months; (2) significant sleep problem lasting at least 3 months, manifested in an average of 30 or more minutes of sleep-onset latency, 30 or more minutes wake after sleep onset, and/or 3 or more awakenings per night based on parent reports; and (3) 2-parent families with both mother and father willing to participate in study procedures. Exclusion criteria were (1) infant pervasive developmental disorder or significant medical illness and (2) any concurrent treatment for infant sleep problems.

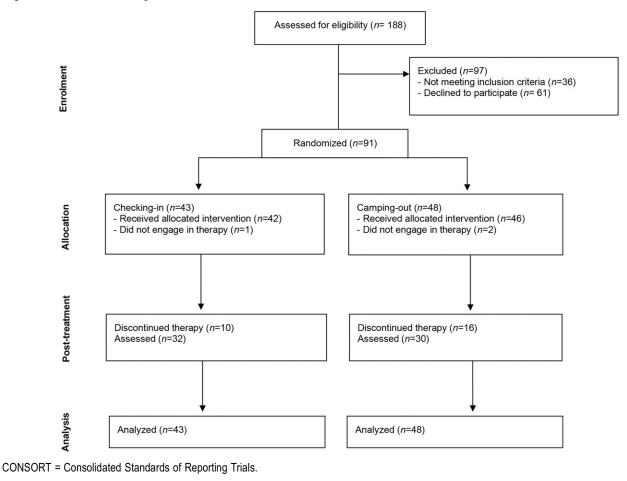
A total of 188 families initially approached the study. Of these, 97 were excluded following the telephone screening interview. The remaining 91 infants (mean age = 12.25 months, SD = 3.11; 56 boys) and their 182 parents were randomized to either Checking-in or Camping-out interventions. Groups did not differ in baseline demographic or clinical characteristics (see **Table 1**). Three families (Checking-in = 1, Campingout=2) did not engage in treatment, and 26 families (Checkingin = 10, Camping-out=16) discontinued treatment or failed to complete the post-treatment assessment. The most prominent reasons for attrition were parent-reported difficulties adhering to the intervention guidelines. There was no significant difference in attrition between groups ($\chi 2[1] = 1.05$, P = .31). Importantly, no differences were found between families who continued and families who discontinued therapy in parent age, years of education, workload, cry tolerance, or sleep-related cognitions, or in infant age, sex, or sleep measures (all P > .19). The Israeli Ministry of Health Institutional Review Board approved the study, and all parents provided written informed consent (Clinicaltrials.gov; Identifier: NCT01489215).

Measures

Actigraphy

Infant sleep—wake patterns were assessed objectively using actigraphy at baseline and post-treatment. Extensive research has established actigraphy as a valid method to assess sleep in

Figure 1—CONSORT diagram.



infants, children, and adults, ^{28–30} and clinical trials have demonstrated the use of actigraphy for documenting the efficacy of sleep interventions, ^{15,31} We used the Micromini actigraph (AMI, Ardsley, NY) in the standard operation mode for sleep—wake scoring. Parents were asked to attach the activity monitor to the infant's ankle for 7 consecutive nights. Sleep diaries were completed by parents and used to detect and amend any irregularities in actigraphic data. Sadeh's algorithm²⁸ was used to obtain the following sleep metrics, based on the inclusion criteria defining the presence and severity of infant sleep problems: (1) minutes of wake after sleep onset and (2) number of awakenings lasting 5 minutes or longer. Actigraphic sleep-onset latency was not used as an outcome measure due to our inability to ascertain lights-out timing.

Brief infant sleep questionnaire

The Brief Infant Sleep Questionnaire (BISQ) is a brief questionnaire used to provide global information on infant sleep as perceived by parents. It has been validated and previously used in numerous studies.^{32,33} The BISQ was completed once by parents together at each assessment point. The measures included in this study were as follows: (1) sleep-onset latency, (2) minutes of wake after sleep onset, (3) number of awakenings, and (4) sleep problem score, representing parental perceived severity of the child's sleep problem.

Intervention delay to infant crying video

PCT was assessed using the Intervention Delay to Infant Crying Video (IDICV).²⁰ Parents were presented with a 2-minute video-clip of a crying infant, with gradually increasing intensity and visual distress signs. Before watching the clip, parents were presented with the following written rationale: "The video you are about to watch is of a very demanding baby. His parents are trying to ignore some of his crying to allow him to calm himself down. Please watch the video and decide when you feel it is absolutely necessary to intervene." This rationale is meant to create a standardized context and to increase parents' motivation to tolerate the crying and delay their response. Parents wore headsets while watching the video, adjusted to an average of 80 db. The delay to intervene (in seconds) was used as the outcome measure, with longer delays representing higher cry tolerance. Both mothers and fathers completed this procedure independently at both assessment points.

The infant sleep vignettes interpretation scale

The Infant Sleep Vignettes Interpretation Scale (ISVIS) was used to assess parental sleep-related cognitions. This questionnaire was developed and validated in previous studies. ^{22,34} It includes 14 hypothetical case descriptions of infants who

Table 1—Demographic and clinical characteristics by group at baseline.

	Checking-in (n = 43)	Camping-out (n = 48)	Between-Group Statistics, t (P)
Child	<u> </u>		
Age, months	12.32 (3.26)	12.11 (2.88)	.32 (.75)
Sex, n (%) male	23 (53)	33 (69)	$\chi^2 = 2.23 (.14)$
Mother			
Age, years	32.00 (3.16)	33.12 (4.05)	-1.42 (.16)
Years of education	16.37 (1.95)	16.40 (1.99)	08 (.94)
Workload, hours/week	31.86 (17.45)	25.78 (16.79)	1.55 (.13)
Father			
Age, years	34.59 (3.97)	35.33 (4.89)	77 (.44)
Years of education	16.05 (2.57)	15.71 (2.56)	.59 (.56)
Workload, hours/week	41.20 (14.79)	43.43 (16.25)	59 (.56)
Actigraphic sleep measures			
Wake after sleep onset, minutes	109.77 (38.61)	119.58 (37.85)	-1.15 (.25)
Number of awakenings	5.86 (1.85)	6.50 (2.27)	-1.38 (.17)
Parent-reported sleep measures			
Sleep-onset latency, minutes	13.59 (13.42)	16.60 (13.96)	-1.01 (.32)
Wake after sleep onset, minutes	64.22 (46.95)	73.08 (58.65)	73 (.47)
Number of awakenings	5.00 (2.32)	4.46 (2.15)	1.10 (.27)
Sleep problem score	4.17 (0.77)	3.88 (0.74)	1.67 (.10)

Data are given as means (SDs) unless otherwise indicated.

display behavioral sleep-related problems, such as difficulties in initiating sleep and recurrent awakenings. After each description parents are asked to rate their agreement with possible interpretations of the described behavior on a 6-point Likert scale. For the purpose of this study, we used the Distress subscale, based on the 14 vignette items, that assesses the extent to which parents attribute distress to infants experiencing sleep disruptions. Higher scores reflect higher distress-attribution and greater endorsement of the importance of parental intervention with soothing the infant at night. Both parents completed the ISVIS at baseline and post-treatment.

Procedures

After an initial phone screening, families meeting the criteria were invited to the university clinic to undergo the baseline assessment procedures. These included laboratory assessments of PCT using the IDICV procedure, 7 consecutive nights of infant sleep home-monitoring using actigraphy, and parental completion of the BISQ, ISVIS, and a demographic background questionnaire. Families were then randomly assigned to 1 of the 2 behavioral sleep intervention groups (ie, Checking-in or Camping-out). At the end of the assessment week, parents were invited to a face-to-face individual session with a clinical psychologist with special training in pediatric sleep and given instructions regarding the intervention. At the middle and end of the week following this session, families were contacted by the psychologist via phone to discuss progress and any concerns or questions. The post-treatment assessment was conducted 1 month following

the intervention and included the same procedures as at baseline, except for the demographic questionnaire, which was not repeated.

Interventions

The Checking-in intervention is based on principles of graduated extinction. Guidelines for this interventions were the following: (1) the infant should be put to bed awake; (2) minimal parental involvement after putting the infant to bed; (3) if the child protests or cries, parents should visit every few minutes (eg, 5 minutes), help the child resume a sleeping position, and find sleep aids (eg, pacifier); (4) parents should then disengage and leave the room until the next visit, even if the child continues with loud protest. Similar rules apply when the child wakes up during the night.

The Camping-out intervention is also extinction based and includes similar schedules as in Checking-in. ^{15,35} In Camping-out, however, the parent is instructed to stay in the room with the infant, remaining present and passive throughout the night. Upon crying or protest the parent intervenes every few minutes to soothe the infant briefly and then returns to the passive sleeping position in a separate bed or mattress next to the child's crib.

Data analysis plan

Independent-samples t and χ^2 tests were used to compare between-group demographic and clinical characteristics at baseline. To test whether PCT and sleep-related cognitions predicted improvement in sleep following treatment, interactions between time and each of these baseline predictors

(PCT and distress-attribution) were modeled using generalized estimating equations (GEEs).36,37 GEEs account for correlated repeated within-subject assessments and accommodate missing data by computing estimated marginal means, which allows for analyses of the intent-to-treat sample. Unstructured covariance matrixes were specified in all models to represent within-subject dependencies. Time (baseline and posttreatment) and parent (mothers and fathers) were entered as within-subject variables in each of the models, and baseline PCT or distress-attribution cognitions were entered as predictors. Significant time-by-baseline predictor interaction terms indicated differential intervention effects as a function of the predictor. Interaction effects were interpreted using post hoc simple slopes analysis, based on a median split allocating participants to low versus high PCT and distress-attribution cognitions (median = 36.00 and 3.69, respectively).³⁸

Treatment effects on PCT and sleep-related cognitions were also evaluated using GEEs. Overall effects of the interventions on PCT and distress-attribution cognitions were estimated using models containing main effects of group (Checking-in and Camping-out), time, and their interaction terms. Finally, to evaluate effect sizes, the standardized mean difference between groups (ie, Cohen's d) was calculated based on the estimated means and standard errors generated from GEE analyses.

To test whether changes in sleep outcomes were associated with changes in PCT and distress-attribution cognitions, missing data were first replaced using the expectation-maximization method, allowing for analysis of data from the entire sample.³⁹ Baseline and post-treatment difference scores were then computed for each variable, and Pearson correlations were used to assess the links between difference scores.

The sample size was determined based on prior studies, which have reported medium to large effect sizes for sleep outcomes (subjective measures usually yielding larger effect sizes compared to actigraphy), 5,17 suggesting that, with a probability level of .05, n = 90 participants would yield 80% power.

RESULTS

Significant improvements in infant sleep were found following both interventions (for a detailed description of these effects, see Kahn et al¹⁹).

Does PCT predict sleep outcomes?

GEE models testing for prediction of baseline PCT (using the IDICV) on reduction in parent-reported sleep yielded significant time-by-cry tolerance interactions for the number of nighttime awakenings (Wald = 7.52, P = .006) and sleep-problem score on the BISQ (Wald = 9.33, P = .002), indicating that cry tolerance predicted changes in reported sleep across treatment groups from baseline to post-treatment. Post hoc analyses revealed a reduction in nighttime awakenings and sleep problems for both low- and high-tolerance parents. However, this improvement was larger for parents with a higher tolerance for crying (for nighttime awakenings: mean difference = 3.93 awakenings, P < .001, Cohen's d = 2.76, 95%

confidence interval [CI] = 2.45, 3.08; for the sleep-problem score: mean difference = 1.83, P < .001, Cohen's d = 1.83, 95% CI = 1.64, 2.03) compared with parents with lower cry tolerance (for nighttime awakenings: mean difference = 3.21 awakenings, P = .001, Cohen's d = 1.84, 95% CI = 1.46, 2.22; for the sleep-problem score: mean difference = 1.55, P < .001, Cohen's d = 1.72, 95% CI = 1.53, 1.92). GEE models testing prediction of changes in parent-reported sleep-onset latency and wake after sleep onset, as well as objectively measured sleep, were nonsignificant.

Do parental distress-attribution cognitions predict sleep outcomes?

Baseline ISVIS distress-attribution scores significantly predicted reductions in parent-reported sleep problems, as indexed by a time-by-distress-attribution interaction (Wald = 6.31, P = 0.01). Simple slopes analyses post hoc demonstrated larger decreases in the infant sleep problems score for parents with lower initial distress-attribution (mean difference = 1.74, P < .001, Cohen's d = 1.76, 95% CI = 1.55, 1.97) compared with parents with higher distress-attribution scores (mean difference = 1.56, P < .001, Cohen's d = 1.64, 95% CI = 1.43, 1.86). The ISVIS distress-attribution scale did not significantly predict changes in other sleep measures following the intervention.

Do behavioral interventions enhance PCT?

A main effect of time was found for PCT using the IDICV score (Wald = 4.93, b = 8.29, SE = 3.73, P = 0.026), indicating that parents became significantly more tolerant to infant crying from baseline to post-treatment (see **Figure 2**). The main effect for group and time-by-group interaction effect were nonsignificant, suggesting that the increase in cry tolerance occurred regardless of intervention group.

Do behavioral interventions reduce distressattribution cognitions?

GEE models for the distress scale on the ISVIS revealed a main effect of time (Wald = 43.74, b = .71, SE = .11, P < .001), indexing a significant decrease in distress-attribution cognitions from baseline to post-treatment (see **Figure 3**). Moreover, the time-by-group interaction effect was significant (Wald = 10.19, b = -.65, SE = .20, P = .001). Post hoc analyses showed that, while the reduction in distress-attribution was significant in both intervention groups, this effect was stronger in the Checking-in group (mean difference = 1.00, P < .001, Cohen's d = 1.62, 95% CI = 1.48, 1.75) compared with the Camping-out group (mean difference = .35, P = .015, Cohen's d = .50, 95% CI = .35, .65).

Are changes in sleep associated with changes in PCT and distress-attribution cognitions?

Improvement in sleep from baseline to post-treatment was associated with increased parental ability to tolerate crying and distress following treatment. Decreases in the reported number of nighttime awakenings and sleep-onset latency, as well as with decreases in actigraphic wake after sleep onset, were significantly associated with increases in PCT (r = -.41, P < .001;

Figure 2—Parental cry tolerance at baseline and post-treatment in the Checking-in and Camping-out groups.

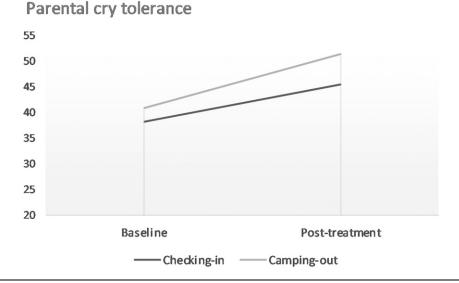
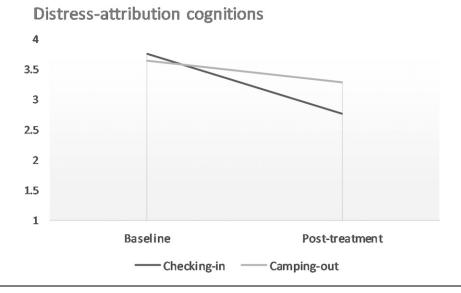


Figure 3—Distress-attribution cognitions scores at baseline and post-treatment in the Checking-in and Camping-out groups.



r=-.24, P=.02; r=-.23, P=.03, respectively, for the different sleep variables). In addition, decreases in parent-reported wake after sleep onset were significantly linked with decreases in distress-attribution cognitions (r=.29, P=.008).

DISCUSSION

This study found that parental tolerance for infant distress predicted improvement in sleep following behavioral treatment for pediatric insomnia. Both higher PCT and lower parental distress-attribution cognitions at baseline predicted greater improvement in parent-reported sleep problems at post-treatment, and higher PCT additionally predicted larger reductions in the number of reported nighttime awakenings. Behavioral interventions for infant sleep-related problems require parents to modify their involvement in the sleep context,

so that infant self-soothing is practiced and learned. ¹² Throughout this process, they must refrain from providing immediate comfort, but rather tolerate some extent of discomfort while delaying their response. Thus, parents who were more highly sensitive may have been less able to effectively implement the intervention, resulting in more modest treatment effects. The results of the current study dovetail with our previous report, demonstrating that heightened infant separation anxiety predicted smaller improvement in sleep following these interventions. ¹⁹ Taken together, these findings represent a novel endeavor to identify infant and parent factors that predict outcomes of behavioral treatments for infant sleep problems.

The present findings correspond with evidence about the predictive role of parenting tendencies in anxiety disorders. Parental accommodation, a pattern of "rescuing" behavior parents tend to display upon signs of child distress, has not

only been linked to symptom severity in anxiety disorders but also to treatment failure. 40,41 Moreover, parent training directed at reducing parental accommodation led to consistent and enduring improvements in the child's symptoms. 42 Hence, in both the anxiety and sleep contexts, parental overinvolvement and low tolerance for distress seem to impede the development of child coping skills, rendering the child dependent on parental regulation. This commonality between the role of parental accommodation in sleep disturbances and anxiety disorders may perhaps indicate an overlap or continuity between the two. This might have implications for prevention and treatment.

Changes in actigraphic sleep measures were not predicted by PCT or distress-attribution in this study, nor were improvements in parent-reported sleep-onset latency and wakefulness after sleep onset. It is not surprising that the sleep metric most consistently predicted by parent factors was the parent-reported sleep problem score. This score reflects parents' general perception of their child's sleep problem severity. Parental cognitions have been shown to shape parents' perception of the gravity (or even existence) of a sleep problem—in some cases even more than the child's specific sleep variables. 26,43 Furthermore, whereas improvements in both objective and reported infant sleep were documented as a result of these interventions, effects were stronger and more robust for parent-reported compared with actigraphic sleep. 19 Conjointly, these findings suggest that some of the most prominent intervention outcomes occur within the parents themselves.

This study found enhancements in parental tolerance for infant distress following the interventions. PCT increased and distress-attribution decreased from baseline to post-treatment, indicating that parents were less likely to interpret sleep difficulties as a sign of distress that requires immediate response. Interestingly, baseline and post-treatment levels of PCT and distress-attribution were equivalent to levels found in parents of infants with and without sleep problems, respectively, in Sadeh and colleagues' cross-sectional investigation.²⁰ These findings are in accord with Hall and colleagues' report of improvement in sleep-related cognitions following a group behavioral intervention for parents of infants with sleep problems. 16 The decrease in distress-attribution in the present study was stronger in the Checking-in compared with the Camping-out group, presumably since parents in this group had to practice distancing themselves from the distressed child, which may have led to greater cognitive changes in the way this distress is perceived.

Interestingly, while the interventions used in this study targeted parental behavior, with only a brief indication of the rationale, effects were not observed solely in the behavioral domain but also in how parents perceived they should cope with infant crying and sleep. It may be assumed that developments in parental tolerance for distress allowed parents to implement the intervention in a better way, creating a positive feedback loop. Their presumed ability to withstand the infant's crying may have led to quicker acquisition of infant self-soothing, resulting in improved sleep for both infant and parents, and in turn to greater parental tolerance for distress. While it is impossible to confirm these assumptions based on the present data, the

associations found between improvement in infant sleep and changes in PCT and distress-attribution following treatment demonstrate the intertwined relations between parenting factors and infant sleep. These findings are in line with Sadeh and Anders' classic transactional model of infant sleep, positing that parent characteristics play a key role in infant sleep processes.⁴⁴

Several limitations merit consideration. First, although the present sample size is large compared with previous infant sleep intervention studies, it might still lack sufficient power to detect some of the expected effects. The sample was also fairly homogeneous and included only families with both a mother and father. Together with the relatively high proportion of dropout (albeit equivalent across treatment groups and unrelated to baseline characteristics), these elements limit the generalizability of our findings. More heterogeneous samples should be used to examine these effects in future investigations. In addition, as a waitlist control was not included, it could be argued that changes in PCT and distress-attribution following treatment were due to the passage of time or other nonspecific effects. Future studies may wish to evaluate the effects of treatment on these constructs compared with an inactive control. Finally, PCT and distress-attribution were not assessed midtreatment; thus, we are unable to ascertain whether these factors mediate treatment outcomes (ie, whether increases in PCT and decreases in distress-attribution precede and are associated with greater improvement in infant sleep). Future investigations should measure these constructs not only following, but also during, the process of treatment to determine whether they are part of the mechanisms underlying the effects of behavioral interventions for pediatric insomnia.

Notwithstanding these limitations, to the best of our knowledge, this is the first study to examine the role parenting factors play in behavioral interventions for pediatric insomnia. As parents are the ones who implement these treatments, identifying parental characteristics that predict outcomes may be of significant clinical value. Our findings inform health-care providers, suggesting which parent populations may require more intensive professional support. They also shed light on the ways in which behavioral interventions affect parents' tolerance for infant distress, implying that these may be some of the "active components" by which these treatments exercise their benefits. Future efforts should be made to design and evaluate "add-on" treatment components targeting low PCT and high distress-attribution cognitions directly, so as to improve outcomes for those parents who may find implementation of these treatments most challenging, despite being the ones that need them the most. A broader implication has to do with the possible continuity between sleep and anxiety disorders. Such a link would make interventions that successfully address the difficulties of parents to "de-accommodate",45 potentially relevant for engaging parents with low PCT and high distress-attribution in treatments for infant sleep problems.

ABBREVIATIONS

BISQ, Brief Infant Sleep Questionnaire CI, confidence interval

IDICV, Intervention Delay to Infant Crying Video ISVIS, Infant Sleep Vignettes Interpretation Scale GEE, generalized estimating equation PCT, parental cry tolerance

REFERENCES

- Jenni OG, LeBourgeois MK. Understanding sleep—wake behavior and sleep disorders in children: the value of a model. Curr Opin Psychiatry. 2006;19(3):282–287.
- Galland BC, Taylor BJ, Elder DE, Herbison P. Normal sleep patterns in infants and children: a systematic review of observational studies. Sleep Med Rev. 2012;16(3):213–222.
- Field T. Infant sleep problems and interventions: a review. Infant Behav Dev. 2017;47:40–53.
- Honaker SM, Meltzer L. Sleep in pediatric primary care: a review of the literature. Sleep Med Rev. 2016;25:31–39.
- Mindell JA, Kuhn B, Lewin DS, Meltzer LJ, Sadeh A. Behavioral treatment of bedtime problems and night wakings in infants and young children. Sleep. 2006;29(10):1263–1276.
- Sadeh A, Tikotzky L, Kahn M. Sleep in infancy and childhood: implications for emotional and behavioral difficulties in adolescence and beyond. *Curr Opi Psychiatry*. 2014;27(6):453–459.
- Mindell JA, Leichman ES, DuMond C, Sadeh A. Sleep and social-emotional development in infants and toddlers. J Clin Child Adolesc Psychol. 2017;46(2):236–246.
- Martin J, Hiscock H, Hardy P, Davey B, Wake M. Adverse associations of infant and child sleep problems and parent health: an Australian population study. *Pediatrics*. 2007;119(5):947–955.
- Bayer JK, Hiscock H, Hampton A, Wake M. Sleep problems in young infants and maternal mental and physical health. J Paediatr Child Health. 2007; 43(1-2):66–73.
- Fairbrother N, Woody SR. New mothers' thoughts of harm related to the newborn. Arch Women Ment Health. 2008;11(3):221–229.
- Wake M, Morton-Allen E, Poulakis Z, Hiscock H, Gallagher S, Oberklaid F. Prevalence, stability, and outcomes of cry-fuss and sleep problems in the first 2 years of life: prospective community-based study. *Pediatrics*. 2006;117(3):836–842.
- Meltzer LJ, Mindell JA. Systematic review and meta-analysis of behavioral interventions for pediatric insomnia. J Pediatr Psychol. 2014;39(8):932–948.
- Burnham MM, Goodlin-Jones BL, Gaylor EE, Anders TF. Nighttime sleep-wake patterns and self-soothing from birth to one year of age: a longitudinal intervention study. J Child Psychol Psychiatry. 2002;43(6):713–725.
- Hiscock H, Bayer JK, Hampton A, Ukoumunne OC, Wake M. Long-term mother and child mental health effects of a population-based infant sleep intervention: cluster-randomized, controlled trial. *Pediatrics*. 2008;122(3):e621–e627.
- Sadeh A. Assessment of interventions for infant night waking—parental reports and activity-based home monitoring. J Consult Clin Psychol. 1994;62(1):63–68.
- Hall WA, Hutton E, Brant RF, et al. A randomized controlled trial of an intervention for infants' behavioral sleep problems. BMC Pediatr. 2015;15(1):181.
- Gradisar M, Jackson K, Spurrier NJ, et al. Behavioral interventions for infant sleep problems: a randomized controlled trial. *Pediatrics*. 2016;137(6):e20151486.
- Price A, Wake M, Ukoumunne OC, Hiscock H. Five-year follow-up of harms and benefits of behavioral infant sleep intervention: randomized trial. *Pediatrics*. 2012;130(4):643–651.
- Kahn M, Juda-Hanael M, Livne-Karp E, Tikotzky L, Anders TF, Sadeh A. Behavioral interventions for pediatric insomnia: one treatment may not fit all. Sleep. 2019;zsz268.

- Sadeh A, Juda-Hanael M, Livne-Karp E, et al. Low parental tolerance for infant crying: an underlying factor in infant sleep problems? J Sleep Res. 2016;25(5):501–507.
- Kahn M, Bauminger Y, Volkovich E, Meiri G, Sadeh A, Tikotzky L. Links between infant sleep and parental tolerance for infant crying: longitudinal assessment from pregnancy through 6months postpartum. Sleep Med. 2018;50: 72–78
- Tikotzky L, Sadeh A. Maternal sleep-related cognitions and infant sleep: a longitudinal study from pregnancy through the 1st year. *Child Dev*. 2009;80(3):860–874.
- Kraemer HC, Frank E, Kupfer DJ. Moderators of treatment outcomes: clinical, research, and policy importance. *JAMA*. 2006;296(10):1286–1289.
- Keeton CP, Ginsburg GS, Drake KL, et al. Benefits of child-focused anxiety treatments for parents and family functioning. *Depress Anxiety*. 2013;30(9):865–872.
- Tarver J, Palmer M, Webb S, et al. Child and parent outcomes following parent interventions for child emotional and behavioral problems in autism spectrum disorders: a systematic review and meta-analysis. *Autism*. 2019;23(7):1630–1644.
- Lebowitz ER, Marin C, Martino A, Shimshoni Y, Silverman WK. Parent-based treatment as efficacious as cognitive-behavioral therapy for childhood anxiety: a randomized noninferiority study of supportive parenting for anxious childhood emotions. J Am Acad Child Adolesc Psychiatry. 2020;59(3): 362–372.
- Hiscock H, Wake M. Randomised controlled trial of behavioural infant sleep intervention to improve infant sleep and maternal mood. *BMJ*. 2002;324(7345):1062.
- Sadeh A. The role and validity of actigraphy in sleep medicine: an update. Sleep Med Rev. 2011;15(4):259–267.
- Sadeh A. Evaluating night wakings in sleep-disturbed infants: a methodological study of parental reports and actigraphy. Sleep. 1996;19(10):757–762.
- Meltzer LJ, Montgomery-Downs HE, Insana SP, Walsh CM. Use of actigraphy for assessment in pediatric sleep research. Sleep Med Rev. 2012;16(5):463–475.
- Kahn M, Ronen A, Apter A, Sadeh A. Cognitive–behavioral versus non-directive therapy for preschoolers with severe nighttime fears and sleep-related problems. *Sleep Med*. 2017;32:40–47.
- Sadeh A. A brief screening questionnaire for infant sleep problems: validation and findings for an Internet sample. *Pediatrics*. 2004;113(6): e570–e577.
- Tikotzky L, Volkovich E. Infant nocturnal wakefulness: a longitudinal study comparing three sleep assessment methods. Sleep. 2018;42(1):zsy191.
- Sadeh A, Flint-Ofir E, Tirosh T, Tikotzky L. Infant sleep and parental sleeprelated cognitions. J Fam Psychol. 2007;21(1):74–87.
- Lam P, Hiscock H, Wake M. Outcomes of infant sleep problems: a longitudinal study of sleep, behavior, and maternal well-being. *Pediatrics*. 2003;111(3):e203–e207.
- 36. Zeger SL, Liang K-Y. Longitudinal data analysis for discrete and continuous outcomes. *Biometrics*. 1986;42(1):121–130.
- Zeger SL, Liang K-Y, Albert PS. Models for longitudinal data: a generalized estimating equation approach. *Biometrics*. 1988;44(4):1049–1060.
- Preacher KJ, Curran PJ, Bauer D. Computational tools for probing interactions in multiple linear regression, multilevel modeling, and latent curve analysis. *J Educ Behav Stat*. 2006;31(4):437–448.
- Pigott TD. A review of methods for missing data. Educ Res Eval. 2001;7(4):353–383.
- Lebowitz ER, Panza KE, Bloch MH. Family accommodation in obsessivecompulsive and anxiety disorders: a five-year update. Expert Rev Neurother. 2016;16(1):45–53.
- Storch EA, Salloum A, Johnco C, et al. Phenomenology and clinical correlates of family accommodation in pediatric anxiety disorders. J Anxiety Disord. 2015;35: 75–81
- Lebowitz ER, Omer H, Hermes H, Scahill L. Parent training for childhood anxiety disorders: the SPACE program. *Cognit Behav Pract.* 2014;21(4): 456–469.

- Sadeh A, Mindell J, Rivera L. "My child has a sleep problem": a cross-cultural comparison of parental definitions. Sleep Med. 2011;12(5):478–482.
- Sadeh A, Anders TF. Infant sleep problems: origins, assessment, interventions. Infant Ment Health J. 1993;14(1):17–34.
- Lebowitz ER, Omer H. Treating childhood and adolescent anxiety: a guide for caregivers. Hoboken, NJ: John Wiley & Sons; 2013.

ACKNOWLEDGMENTS

The authors thank the participating families and team of research assistants. They dedicate this paper to their dear colleague, Professor Avi Sadeh, who led this study, and devoted his career to support families in their pursuit of sound sleep.

SUBMISSION & CORRESPONDENCE INFORMATION

Submitted for publication January 7, 2020 Submitted in final revised form April 2, 2020 Accepted for publication April 3, 2020

Address correspondence to: Michal Kahn, PhD, School of Psychological Sciences, Tel-Aviv University, Tel-Aviv 69978, Israel; Email: michalkahn10@gmail.com

DISCLOSURE STATEMENT

All authors have seen and approved the manuscript. Work for this study was performed at Tel Aviv University. This study was funded by the United States—Israel Binational Science Foundation (BSF) (grant 2009229). The authors report no conflicts of interest.