

#### SCIENTIFIC INVESTIGATIONS

# A brief behavioral treatment for unresolved insomnia in adolescents: a single-case multiple baseline pilot study, evaluating self-reported outcomes of efficacy, safety, and acceptability

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Study Objectives: Insomnia is a significant problem for many adolescents and often is associated with detrimental effects on physical and mental health. Drawing on emerging models of intervention in the adult literature, this pilot study investigated the efficacy, safety, and acceptability of a novel, brief (3-week) behavioral intervention among adolescents with unresolved chronic insomnia.

Methods: A multiple baseline (staggered start) A-B with follow-up single-case design (n = 2) was used to evaluate intervention outcomes across treatment and at 2-month follow-up in the domains of sleep, mood and affect, fatigue, and parent-child conflict. Outcomes were indexed with multi-informant data collected using adolescent reports on mood/affect and sleep diaries, and parent reports on parent-child conflict.

**Results:** Posttreatment and 2-month follow-up data indicated improvements in self-reported sleep quality, including sleep onset latency and increased sleep efficiency. Indicators related to participant mood, stress, and parent-child interactions remained relatively stable over the course of treatment, suggesting that the sleep restriction component of the intervention did not produce adverse effects for the adolescents or their families.

**Conclusions:** A brief 3-week intervention adapted from the adult literature was associated with improved sleep-wake cycles in adolescents with chronic insomnia. Change during the treatment phase was particularly rapid and maintained over time. In conjunction with low observed risk and adverse effects, the potential for this treatment to provide a safe, acceptable, and cost-effective manualized treatment for adolescent insomnia warrants larger-scale clinical evaluation. **Clinical Trial Registration:** Registry: Australian New Zealand Clinical Trials Registry; Title: Does a one month brief behavioral treatment improve sleep for high school adolescents (ages 12-17): an open label pilot study; Identifier: ACTRN12618000835246; URL: https://www.anzctr.org.au/Trial/Registration/TrialReview.aspx?id=375102

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#### BRIEF SUMMARY

**Current Knowledge/Study Rationale:** Brief behavioral interventions are effective in the treatment of adult insomnia, yet their effectiveness among adolescents is less understood; moreover, it is unknown whether the sleep restriction component of such interventions may produce adverse effects in this developmental period. Consistent with the push for briefer cognitive behavior therapy for insomnia treatments, we adapted a brief behavioral treatment from the adult literature and evaluated whether this intervention can be delivered efficaciously and safely to adolescents.

Study Impact: A novel, brief (3-week) behavioral intervention was associated with improved self-reported sleep quality posttreatment and at 2-month followup. Moreover, it was found to be safe and highly acceptable to participants. The intensive case-series data collected in this study indicate that this manualized intervention warrants further evaluation in large-scale controlled clinical trials focused on cost-effectiveness and dissemination in various adolescent populations.

#### INTRODUCTION

Insomnia is a significant problem for adults as well as children and adolescents. In Western countries, approximately 10% of adolescents meet Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) criteria for the disorder, and a third experience significant symptoms of insomnia.<sup>1–3</sup> Given the known effects of insomnia on physical and mental health, and social and emotional well-being,<sup>4,5</sup> the potential cost burden of adolescent insomnia is extensive. Relatively little attention, however, has focused on treatment during adolescence compared to adulthood and childhood.<sup>6</sup> Two prevailing treatment modalities persist for insomnia, namely pharmacotherapy and cognitive behavior therapy (CBT). Although it is understood that pharmacological interventions for insomnia are at times necessary,<sup>7,8</sup> concerns have been raised regarding the efficacy and safety of drugs commonly prescribed for insomnia at young ages, and treatments that do not involve medication are recognized as far preferable.<sup>9–13</sup> With respect to nonpharmacological intervention, CBT for insomnia (often referred to as CBT-I), is recognized as an effective frontline treatment for adults as well as adolesents.<sup>6,9,14</sup> A full course of CBT-I, however, is often expensive and time consuming.<sup>15–18</sup> Moreover, CBT-I involves significant motivational demands that may act as a barrier to treatment<sup>6</sup> and there appear to be insufficient therapists equipped to meet the need for this individual therapy at the population level.<sup>17</sup>

These challenges have spurred interest in the development of more accessible and cost-effective models of CBT-I, as seen in self-help CBT-I,<sup>19</sup> modified CBT-I,<sup>20</sup> digital CBT-I,<sup>21</sup> computerized CBT-I,<sup>22</sup> and group and Internet CBT-I.<sup>23</sup> Another approach has been to limit CBT-I programs to behavioral components of CBT-I that typically target processes including sleep hygiene,<sup>24</sup> stimulus control,<sup>25</sup> and sleep restriction.<sup>26,27</sup> A recent review of such interventions emphasized promising effects in pediatric/adolescent populations,<sup>28</sup> yet these interventions are often still lengthy and intensive to administer, as emphasized in literature addressing barriers to the implementation of these behavioral approaches.<sup>29</sup> Zhou and Owens suggested that key challenges include engaging adolescents in treatment,<sup>28</sup> whereas Boerner et al highlighted the need for briefer intervention materials.<sup>29</sup>

Interest in brief behavioral interventions for insomnia has likewise grown,<sup>15,30–32</sup> in part to address challenges associated with motivation and engagement with treatment programs that are lengthy and intensive.<sup>33</sup> As a case in point, Buysse et al and Troxel et al developed a brief behavioral intervention for insomnia for older adults (brief behavioral treatment for insomnia [BBT-I]).<sup>15,30</sup> This intervention is delivered over a condensed timeframe of 4 weeks and is restricted to four basic principles from the sleep literature,<sup>26</sup> including psychoeducation on sleep hygiene and stimulus control, in conjunction with sleep restriction principles (eg, limiting time in bed [TIB] and going to bed only when sleepy, not tired). BBT-I has demonstrated efficacy among older adults<sup>15,30,31</sup> and we expect this brief adult intervention may also be well suited to adolescents. In addition, given known barriers for the treatment of insomnia in the adolescent age group,<sup>33,34</sup> we expected that relatively simple changes during the research process might help build the therapeutic alliance and influence teenagers "buy-in" to the program and thus their treatment outcomes. Specifically, we hypothesized that inviting adolescents to engage in a sleep efficiency enhancement program and collaborating with them to set limits on their sleep routines might be a more attractive option than a sleep restriction program that is prescribed by the researcher/therapist. The benefits of a BBT-I adapted for adolescents include potentially providing an earlier entry point to the rapeutic intervention in a stepped care model and reducing the level of training required for practitioners delivering such interventions. If effective, a brief behavioral intervention could therefore be of considerable value to the large number of youths who currently experience the negative effects of insomnia, yet whether such an approach is suitable for adolescents remains untested.

Given the unique developmental characteristics of the adolescent period, caution is required when adapting interventions originally designed for adult populations. Importantly, research has shown that restricting sleep among healthy adolescents can have negative effects on mood and emotion regulation, including increased anxiety, hostility, confusion and fatigue,<sup>4</sup> as well as reduced cognitive function and emotion dysregulation.<sup>4,5</sup> Given that adolescents experiencing insomnia already report less sleep, it is reasonable to question whether further impositions or restrictions to their sleep or opportunities to sleep (eg, via reduced time in bed), could have detrimental effects. It is therefore important that safety is examined as part of evaluations concerning novel interventions for adolescents, particularly when such interventions are designed to affect sleep-wake cycles.

The aim of the current study was to evaluate the efficacy, safety and acceptability of a brief behavioral treatment for chronic unresolved insomnia for adolescents. It was hypothesized that this brief intervention would be associated with significant improvements in scores on the Sleep Condition Indicator (SCI; indicating improved sleep outcomes),<sup>35</sup> and that these improvements would be maintained at 2-month followup. With respect to safety, we examined potential adverse effects across three domains, namely, mood (eg, depression, anxiety, and stress), social relationships (eg, parent-child conflict) and daytime fatigue. Specifically, we evaluated the null hypothesis of no change in scores on the depression, anxiety, and stress scale for youth,<sup>36</sup> on the Flinders fatigue scale,<sup>37</sup> and on the parent-child conflict scale.<sup>38</sup> With respect to acceptability, we examined participants' reflections and reports on their experience of, and engagement with, the intervention.

#### METHODS

#### Participants

Participants were recruited as part of clinical trial (ACTRN12618000835246 registered at https://anzctr.org.au). Inclusion criteria were: aged 12 to 17 years (years 7-10 high school), capable of providing informed written consent, parental/caregiver consent, literacy in English, symptoms of insomnia disorder<sup>39</sup> as identified by parent/caregiver during telephone screening (ie, difficulty initiating or maintaining sleep or waking up too early for at least 3 nights per week, for at least 3 months, with adequate opportunity and circumstances for sleep and at least one daytime impairment related to the sleep difficulty), SCI scale scores below 20, not previously treated with CBT-I. Exclusion criteria were: existing diagnosis of a severe psychiatric disorder (eg, schizophrenia and other psychoses), sleep disorders other than untreated insomnia (eg, sleep apnea, periodic limb movement disorder), severe cognitive impairment that does not allow participants to consent or follow treatment instructions, recent time-zone travel (within last 1 month). The two adolescents aged 15 to 16 years (one male, one female) were referred by their parents for treatment of chronic insomnia. In both cases this insomnia was unresolved following previous medical evaluation and treatment attempts. Symptoms included lengthy sleep onset latency (SOL), dysregulated sleep patterns, inconsistent sleep-wake cycles, and weekly sleep debts (accumulated weekday fatigue compensated by long weekend sleep-ins). Both participants met DSM-5 criteria for insomnia based on clinical assessment (GOS, registered provisional psychologist) and self-report using the SCI ( $\leq 16$ ).<sup>35</sup> Both participants gave informed consent.

Persephonie (pseudonym) was a 16-year old female in year 10 of high school who identified "wanting to see an improvement in my energy levels and ability to stay asleep at night." She reported a history of insomnia exceeding 2 years. Previous medical assessment had excluded organic causes for her insomnia. Presentation included lengthy SOL regardless of bedtime, difficulty maintaining sleep and falling back to sleep, and intermittent night awakenings. She reported anxiety about sleep during her bedtime routine, and when lying awake in bed, which significantly affected her sleep onset. She reported ongoing treatment with a psychologist for symptoms of anxiety and depression over the past 12 months and that treatment had not specifically targeted her sleep/insomnia. She had trialed melatonin without success for approximately 6 weeks, 10 months prior to the current intervention, as well as herbal remedies including teas and valerian root extract, with minimal effect. She reported no prior CBT interventions for her sleep difficulties.

Phobos (pseudonym) was a 15-year old male in year 10 of high school who identified wanting to "increase the amount of sleep I have." He reported a long-standing history of insomnia beginning in primary school (aged approximately 8 years). He reported a concurrent diagnosis of attention deficit hyperactivity disorder. Previous medical assessment indicated no other physiologically based causes for insomnia although some sleep difficulties appear to have coincided with the introduction and trialing of medications in late primary school to early high school (aged 12 to 14 years). He reported taking prescribed melatonin nightly for his sleep difficulties and had few night awakenings, although he identified having difficulty initiating sleep and waking up tired. He also reported racing thoughts prior to sleep onset. He reported no prior CBT interventions for his sleep difficulties.

#### Design

A staggered start single-case A-B design with follow-up was used in this study.<sup>40-42</sup> The staggered-start design is a standard variant of the multiple baseline design.<sup>40</sup> Multiple baseline designs have greater experimental control when compared to simple A-B designs, and are particularly useful when it is impracticable to withdraw treatment.<sup>40,41</sup> For example, in the current study, the effect of the initial treatment precluded a return to baseline, thus excluding A-B-A-... designs. Using a staggered-start, single-case design allowed successive introduction of the intervention thus increasing experimental control because it allowed treatment effectiveness to be evaluated at different times and confounders could be identified systematically relative to treatment start.<sup>41</sup> As the A-B-A design was not appropriate, the current design intentionally replaced the with-drawal phase with follow-up instead to evaluate treatment outcomes.

## Measures

#### Sleep diary

Participants completed self-report sleep diaries each morning for the duration of the study. This included 1-week prior to the intervention (baseline) and 1-week prior to the follow-up consultation. Calculated sleep parameters approximated to the quarter hour included total sleep time (TST), SOL, TIB, sleep efficiency (SE), and night awakenings. Sleep diaries are consistently used in insomnia research, and for adolescents.<sup>43</sup>

#### The Sleep Condition Indicator

The SCI<sup>35</sup> is a reliable and valid clinical screening tool for insomnia. It is consistent with DSM-5 diagnostic criteria. It contains eight items scored from 0 to 4, with higher scores indicative of better sleep (eg, "how long does it take you to fall asleep?" 0 to15 minutes [4], ...,  $\geq$  61 minutes [0]). Scores range from 0 to 32, with a cutoff score  $\leq$  16 indicative of probable insomnia disorder.

#### The Depression, Anxiety and Stress Scale, Youth Version

The Depression, Anxiety and Stress Scale, Youth Version  $(DASS-Y)^{36}$  is a 24-item questionnaire assessing depression, anxiety and stress among youth. Adapted from the DASS,<sup>44</sup> items are rated on a 4-point Likert scale (0 = not true of you, 3 = very true of you), with scores ranging from 0–24 on each of the three subscales. Interpretation of the DASS-Y is similar to the DASS, and the DASS-Y has demonstrated good internal reliability and construct validity.

#### The Flinders Fatigue Scale

The Flinders Fatigue Scale (FFS)<sup>37</sup> is a 7-item scale that assessed fatigue, the "extent to which you have felt tired, weary, exhausted, over the last two weeks." Six of the items are scored on a 5-point scale (0 = not at all, 4 = extremely), with one item assessing when fatigue is experienced (eg, early morning through to late evening), across seven time-points. Total FFS scores are summed and range from 0 to 31. The scale has good internal reliability ( $\alpha = .86$ ), discriminant validity, and clinical utility.

#### The Conflict Behavior Questionnaire

The Conflict Behavior Questionnaire (CBQ-20) is a 20-item true/false questionnaire that assesses relationship quality and conflict between adolescents and their parent(s). Participants completed the questionnaire separately for each parent. The attending parent also completed the questionnaire. Items endorsing distress/conflict were summed thus providing three scale scores—two adolescent-parental conflict scores (adolescent's perceived relationship distress with attending and nonattending parent) and one parental-adolescent conflict score (attending parent's perceived relationship distress with adolescent). Robin and Foster<sup>38</sup> provide normative data for the CBQ-20 and report that it has good internal reliability ( $\alpha = .90$ ) and evidence to support construct validity.

#### Treatment evaluation measure

At treatment end, participants and their parent(s) were asked four open-ended questions: What was the best and worst thing about the treatment? What would I change to make it better? At the beginning of the study, I hoped that (...). They were also asked to rate seven items using a 7-point Likert scale. Items assessed if they were likely to continue to use the sleep program, whether the sleep program changed their sleep quality and quantity and improved understanding of their sleep/wake cycles, if they felt that they could and would use learned strategies to better manage their sleep, and the extent to which the intervention met expectations.

#### Procedure

The University of Sydney's Human Research Ethics Committee approved this study, protocol number: 2018/091. Parents of the adolescents responded to community advertising by emailing the trial coordinator, a provisionally registered psychologist (GQS) supervised by a senior clinical psychologist (DH), who arranged a phone interview with both parent and adolescent in attendance. Participants satisfying inclusion criteria then received electronic copies of the sleep diaries with instructions for completion and scheduled an appointment to attend the research clinic after 7 nights. Both participants attended a face-to-face consultation on the eighth day accompanied by their parent, where they provided written consent and completed baseline questionnaires (eg, the SCI, DASS-Y, FFS, and the CBQ-20).

Participants used sleep diaries to record sleep patterns at home. Participants and their attending parent completed paper and pencil questionnaires while attending the University of Sydney for baseline, treatment-end and follow-up appointments. Baseline data consisted of questionnaires completed at the first appointment and sleep diary records for the 7 days preceding that appointment. Verbal responses were transcribed during the two 15-minute midintervention phone calls, which included assessing and modifying sleep routines with participants and discussing any changes with parents. Participants continued to record their sleep behavior throughout treatment and for the 7 days preceding 2-month follow-up.

Treatment consisted of a 1.5-hour information and planning session based on a modified version of the BBT- $I^{15,30}$  (session 1) and two 10- to 15-minute phone calls each 1 week apart (sessions 2 and 3). Treatment was evaluated at treatment end (session 4) and at 2-month follow-up (session 5). Session 1 consisted of a question-and-answer-style information session on sleep hygiene, stimulus control, and sleep-wake cycles developed specifically for adolescents. The education component was designed to engage participants by identifying and discussing biological sleep drivers (eg, why and how much), adolescent sleep cycles, and the costs and benefits of changing their sleep patterns with reference to behavioral, cognitive, physical, and social functioning specific to their identified needs. With participant 'buy-in', session 1 then focused on learning and applying the four principles of the BBT-I as specified in the manualized program for adults (ie, reduce TIB, get up at the same time each day, go to bed only when sleepy, get out of bed if not asleep)<sup>30</sup> and setting appropriate bedtimes and rise times. Importantly, to regulate sleep patterns, participants and their parent identified an appropriate rise time for the duration of the intervention. Using their baseline sleep diary data, the treatment team (adolescent, parent, and coordinator) calculated then negotiated a suitable sleep-wake cycle. In the adult literature, the set bedtime is TST plus 15 to 30 minutes, with minimum TIB usually approximately 5 hours. Vital to our approach, adolescents were empowered in the decision-making process because we thought it important that our participants engage with the program by being "able to live with it." We therefore used a less restrictive approach as the starting point

from which to negotiate, as we hypothesized that this would enhance acceptability and thus treatment through improved adherence. This included informing participants of a minimum TIB of 6 hours, then calculating TST and negotiating bedtimes and rise times acceptable to the treatment team. For Persephonie, this involved an initial bedtime of 1:30 AM and a rise time of 8:00 AM, with TIB 6.5 hours, and for Phobos, an initial bedtime of 10:45 PM and a rise time of 6:00 AM, with TIB 7.25 hours. Over the course of treatment we increased TIB by a negotiated 15 or 30 minutes during discussion in the midtreatment phone calls (sessions 2 and 3) if SOL was less than 30 minutes and SE was greater than 85% (compared with the more rigid approach of 15 minutes when  $SE \ge 90\%$ ,<sup>26</sup> and decreased TIB if SOL was greater than 30 minutes. Participants maintained their sleep diaries throughout treatment to facilitate weekly adjustment to their sleep times. Consultation and decision making with the adolescent, not for the adolescent, was vital in our procedure.

#### Statistical analyses

Differences in baseline and follow-up scores on the SCI were evaluated using the reliable change index (RCI) calculated by Espie et al (six points provides evidence of clinically and statistically significant change).<sup>45</sup> For the FFS, the calculated RCI using the original psychometric data<sup>37</sup> was 5.39. Youden J index of discriminability also provided useful clinical cutoff ratings.<sup>46</sup>

## RESULTS

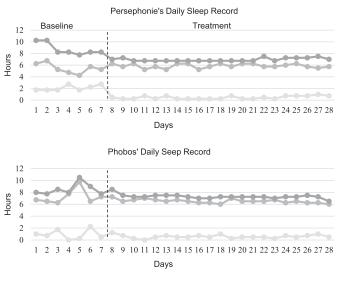
Demonstrable change was evident for both participants over the course of treatment (Figure 1). Visual inspection of the individual sleep records showed change in the variability and differential between TST and TIB, and SOL, preintervention and postintervention. A measure of SE, the differential between TST and TIB was consistently smaller from around the start of treatment (day eight), whereas SOL appeared consistently more stable and shorter for both participants, although more obviously for Persephonie. This narrowing of the gap between TST and TIB (and reduced SOL) suggested improved SE and general adherence to the intervention, with no obvious spikes in TIB on the weekends (sleep-ins are a common pattern associated with adolescent sleep-debt cycles). Based on visual inspection (Figure 1 and Table 1), the effect of the intervention for TST differed for the two participants, though indicated sleep quality improved more than sleep quantity.

#### **Treatment effects**

#### Participant 1: Persephonie

Cores for sleep efficiency (TST  $\div$  TIB) and the sleep condition indicator are reported in **Table 1**. For Persephonie, TST increased by 21 minutes and TIB decreased by 1.54 hours from baseline to treatment end, and her SOL decreased by 1.44 hours. Her SE changed from 62.6% at baseline to 85% (week 1), 88.6% (week 2) and 80.1% (week three), which coincided with increasing her bedtime by an additional 15 minutes in week 2 and by 30 minutes in week three. Reduced SE in week three suggests TIB was increased too rapidly. Her sleep condition indicator score improved by five points (six points is significant

Figure 1—Individual adolescent sleep data from baseline to treatment end.



Day 1 of Persephonie's sleep diary relates to Friday night to Saturday morning. Day 1 of Phobos' sleep diary relates to Tuesday night to Wednesday morning.

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using the reliable change index)<sup>45</sup> and her sleep became less disrupted. During the 7-day baseline phase, Persephonie experienced six-night awakenings on four separate nights. Night 2 and 5 included a 1-hour awakening and a halfhour awakening; Night 3 included a 1-hour awakening; night 4 included a half-hour awakening. During the 21-day intervention phase, she experienced a single night awakening, which lasted 1 hour.

Assessing safety (**Table 1**), Persephonie's scores on depression remained stable in the moderate-severe range; anxiety was in the extremely severe range, although decreased from baseline to treatment end and at 2-month follow-up; stress remained stable in the extremely severe range<sup>36</sup> throughout treatment and at 2-month follow-up, whereas fatigue was significantly lower in week 1 and at 2-month follow-up, despite remaining in the high range<sup>46</sup> throughout the study. Scores reflecting parent-adolescent conflict suggested no change from baseline to treatment end in either perceived conflict toward parents, or parental perceived conflict. All scores were within one standard deviation of the nondistressed mean.<sup>38</sup>

Assessing acceptability, Persephonie reported that the best thing about the intervention was having "a steady plan to follow—it was easier to follow rules" and the worst that "it was hard to stay awake and out of bed until late" (1.30 AM at commencement). Using a seven-point scale (not at all definitely), she indicated that she would (1) "definitely" continue to use the program, (2) "definitely" felt confident that she could use the strategies to better manage her sleep, and (3) "definitely"

	Baseline	Treatment Week 1	Treatment Week 2	Treatment End	Follow-up
Persephonie					
TST (hours)	5.46	5.82	5.96	5.82	7.04
TIB (hours)	8.75	6.86	6.75	7.21	8.18
SOL (hours)	2.11	.43	.32	.67	.32
SE (%)	62.6	85.0	88.6	80.1	86.0
SCI	2			7	5
Depression	11	10	11	13	9
Anxiety	17	12	11	13	10
Stress	24	21	23	24	21
Fatigue	30	24	26	28	21
Phobos					
TST (hours)	7.25	6.79	6.43	6.36	7.00
TIB (hours)	8.50	7.60	7.20	7.10	7.60
SOL (hours)	0.93	0.57	0.57	0.61	0.36
SCI	10			17	21
SE (%)	85.4	89.9	89.7	89.0	92.1
Depression	0	1	0	0	2
Anxiety	3	3	8	9	7
Stress	7	7	12	10	12
Fatigue	15	16	13	14	13

Depression, anxiety and stress scores from DASS-Y. Fatigue scores from Flinders Fatigue Scale. DASS-Y = Depression, Anxiety and Stress Scale, Youth Version; SCI = Sleep Condition Indicator; SE = sleep efficiency (TST ÷ TIB); SOL = sleep onset latency; TIB = time in bed; TST = total sleep time.

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would continue to use these strategies to better manage her sleep. Using a five-point scale ([0] not at all to [4] completely), she reported that the sleep program had improved her sleep quality and quantity "a lot" (scores of 3) and her understanding of her sleep-wake cycle completely (4). She suggested that "not much" needed to change, except that the sleep diary could have been easier to "understand/fill out." Asked to what extent the intervention met expectations, she reported it "exceeded expectations."

Persephonie's mother completed an equivalent posttreatment questionnaire. She reported that the most positive outcome of the intervention was Persephonie "spending much more time with us in the evenings, rather than just lying in bed," and the worst involved "long, cold nights waiting to go to bed and undoing years of parental response to late nights." She indicated that she would "definitely" continue to encourage use of the intervention, and "definitely" agreed that they could and would continue to use the strategies. She reported that the sleep program had improved Persephonie's sleep quality and quantity "a little" (scores of 2) and that her understanding of her daughter's sleep-wake cycles had improved "a lot" (3). She made no suggestions for change and noted that the intervention had "exceeded expectations" with respect to initial goals ("for Persephonie to get longer, better sleep, and to learn not to take devices to bed and to regard her bed/bedroom as part of a loving self-care routine").

#### Participant 2: Phobos

For Phobos, TST decreased by 53 minutes, TIB decreased by 1.40 hours, and his SOL decreased by 19 minutes from baseline to treatment end. His SE changed from 85.4% at baseline to 89.9% (week 1), 89.7% (week 2) and 89.0% (week 3), which coincided with increasing his bedtime by an additional 15 minutes in week 2 and by 30 minutes in week 3. His SCI score improved by seven points, indicating significant improvement. In addition, his score moved beyond the cutoff point for probable insomnia suggesting diagnostic change.<sup>45</sup> Phobos' sleep remained relatively uninterrupted, with no reports of night awakening during the baseline phase, and one half hour night awakening during treatment, with an additional report that on one occasion he awoke in the chair next to his bed where he had fallen asleep prior to the allocated bedtime, and that this then delayed his sleep onset.

Assessing safety (**Table 1**), Phobos' scores on depression remained stable in the healthy range; anxiety was in the healthy range at baseline and increased to the severe range at treatment end and moderate range at 2-month follow-up; stress was in the healthy range at baseline and increased to the mild range at treatment end and moderate range at 2-month follow-up; stress was in the healthy range at baseline and increased to the mild range at treatment end and moderate range at 2-month follow-up.<sup>36</sup> Fatigue remained stable and in the low to borderline range.<sup>46</sup> With respect to the CBQ-20, Phobos' scores were unchanged and in the nondistressed range from baseline to treatment end and at follow-up. His mother's ratings on the CBQ-20 did vary between baseline (5), treatment end (8), and follow-up (7). Compared to the normative nondistressed mean (mean 2.4, standard deviation 2.8),<sup>38</sup> her CBQ-20 scores were elevated at treatment end and follow-up, although at the lower end of the normative distressed mean (mean 12.4,

standard deviation 5.0). Importantly, these scores did not change significantly.

Assessing acceptability, Phobos reported the best thing about the intervention was that "I could fall asleep in less than an hour regularly" and the worst was "having to wake up at 6:00 AM each day, regardless of the day, and the fatigue that comes with it." Using the same scales as Persephonie, he indicated that he would (1) "probably" continue to use the program, (2) "definitely" felt confident that he could use the strategies to better manage his sleep, and (3) "probably" would continue to use these strategies to better manage his sleep. He reported that the sleep program had improved his sleep quality "a lot," sleep quantity "a little" and his understanding of his sleep-wake cycle "a lot" (scores of 3, 2, and 3, respectively). Asked to what extent the intervention met expectations, he replied somewhere between "a little and a lot."

Phobos' mother reported the best thing about the intervention was that "my son has been empowered, feels he has more control, sleep problems have been de-demonized," and the worst involved "having to get up at 6:00 AM daily to make sure he gets up." She indicated that she would "definitely" continue to encourage use of the intervention and could and would use the strategies. She reported that the sleep program had improved Phobos' sleep quality and quantity "a little" (scores of 2) and that her understanding of her son's sleep-wake cycles had improved "a lot" (3). Suggestions for improvement included introducing treatment sessions at or through the school, and she replied "a little" when asked to what extent the trial had met expectations with respect to the initial goal ("we would improve sleep initiation").

#### Two-month posttreatment follow-up

Data obtained at 2-month posttreatment end follow-up included scores on the SCI, DASS-Y, FFS, and CBQ-20 (Table 1). Persephonie and Phobos also completed the sleep diary record, which provided data for TST, TIB, SE and SOL, for the 7 days preceding follow-up assessment. With respect to the primary hypothesis, Persephonie's sleep condition indicator at follow-up remained higher than baseline (ie, improved sleep quality), though lower than treatment end. With reference to the RCI,<sup>45</sup> both changes were nonsignificant (ie,  $\leq 6$ ), although at follow-up compared with baseline, her SE improved by 23.4%, placing her SE in the healthy range (86%): she recorded less TIB (-34 minutes), yet this coincided with more sleep (an additional 1.58 hours). Phobos recorded a further increase of four points on the SCI, solidifying his significant initial gains relative to baseline assessment. Notable in Phobos' results are the average change in his TIB, TST, and SE. Both TIB and TST were marginally lower at follow-up than at baseline, yet his SOL was markedly improved, changing from 55 minutes to just 22 minutes (Table 1). His SE at follow-up was 92%. In conjunction with changes in SOL, this indicated that Phobos' TST might benefit by increasing his TIB. With respect to safety, collectively, the results presented for both participants suggest few adverse effects associated with the implementation of this brief behavioral intervention (Table 1).

#### DISCUSSION

Two adolescents aged 15 to 16 years with unresolved chronic insomnia were found to show clinical improvements in selfreported sleep-wake cycles in response to a novel 3-week brief behavioral intervention. This included reduced SOL, increased SE, and reduced symptom severity on a measure for DSM-5 defined insomnia. Although sleep quantity remained below that recommended for the respective age group, this was unsurprising given that the intervention prioritized sleep quality over quantity in the short term. Additional improvement on measures of sleep quality and quantity at 2-month follow-up were seen, in conjunction with descriptive data regarding high acceptability. This suggests that the skills acquired by participants during the active phase of the intervention, along with the rapid gains experienced during this time, were enough to motivate and empower the participants to continue implementing the intervention autonomously.

Treatment efficacy and adverse effects were indexed using visual, statistical, and descriptive methods.<sup>47</sup> Visual analysis of data indicated greater consistency in sleep patterns during the intervention phase compared to baseline for both participants. Weekend sleep-ins, a characteristic of poor adolescent sleep,<sup>48</sup> were noticeably absent. Adolescents tend to incur a sleep debt during the week that they pay off by sleeping in on the weekend. However, this inadvertently reinforces negative sleep-wake patterns as it sets them up for poor sleep on the next school night because, having slept in, their sleep need is low. For the two participants, the respective intervention strategies and clinical support appears to have empowered them to break this cycle, a point Phobos commented on directly in the follow-up consultation when he described maintaining his sleep-wake schedule postintervention despite noting at treatment end that getting up at the same time every day "was the worst."

It was considered important to examine potential adverse effects that might be associated with a restrictive sleep intervention in the adolescent period. Four key findings were apparent, suggesting the intervention was safe. First, despite some mood fluctuations, there appeared little evidence to suggest systematic effects. Anxiety and stress increased for Phobos, yet depression remained stable, whereas anxiety decreased for Persephonie, while depression and stress remained stable. These variations suggest something other than the intervention may have affected mood. When questioned, participants themselves attributed mood fluctuations to daily life challenges, not the intervention. Second, based on independent parent and participant reports, perceived interpersonal conflict seemed largely unaffected during the intervention. Third, despite considerably reduced TIB, self-reported fatigue remained relatively stable for Phobos and decreased for Persephonie. Fourth, in contrast to what might have been expected, TST did not vary simply as a function of reduced TIB. Whereas Persephonie's reported sleep followed the expected upward trend throughout the study, Phobos reported a downward trend during the active trial phase, returning near to baseline at 2-month follow-up. Despite this, both participants recorded improved sleep quality as indicated by improved SE, reduced SOL, fewer night awakenings, and improved scores on the SCI. Collectively then, these data suggest that this is a safe and seemingly effective brief intervention ready for larger-scale evaluation.

Despite the intervention producing no noteworthy adverse effects, inconsistent TST changes observed during the active trial phase of the study warrants additional consideration. Phobos recorded an average of 43 minutes less sleep per night, and although higher than baseline, Persephonie's TST remained below 6 hours during this phase, well below recommendations. Given that restricting sleep among healthy adolescents, by even small amounts, was found to detrimentally affect mood and behavior,<sup>4</sup> this might be regarded by some as a serious shortcoming of the current study. However, the current study differed from the study by Baum et al<sup>4</sup> in three important ways. First, adolescents experiencing insomnia already experience less sleep than healthy sleeping adolescents. Thus, the effects of a restrictive sleep intervention might arguably be less for participants with insomnia. Second, our participants also varied such that Phobos began with a relatively stable sleep pattern and much higher sleep efficiency index than Persephonie. Simply stated, Persephonie reported less sleep than Phobos and therefore had more to gain, much like our participants compared with participants in the study by Baum et al. Third, and most important, the current intervention focused on restricting TIB, not sleep. Despite substantial reductions to TIB during the active trial phase, benefits were observed during this phase, at treatment end, and at 2-month follow-up. As such, the results support the efficacy and safety of restricting TIB (as distinct from restricting sleep) for adolescents to enhance sleep outcomes.

Zhou and Owns suggested that a key aim for sleep intervention research is to better understand processes for effectively engaging adolescents in such treatment.<sup>28</sup> We addressed this in two key ways. First, we reframed and renamed sleep restriction therapy (SRT) as sleep efficiency enhancement (SEE) for our participants. Potentially negative perceptions about SRT could act as a barrier to treatment, which we considered an unfortunate by-product of the potentially misleading nomenclature that exists. That is, sleep is not restricted in SRT. Rather, SRT restricts TIB to increase SE and thus sleep quality. We believe SEE is a more accurate description, and importantly, may be more palatable to adolescents. Accordingly, rather than focusing on restrictions, participants were presented with material regarding the benefits of increasing their SE and bringing into line their TIB and TST to reduce SOL and to reduce time for rumination and anxiety around not falling asleep. Second, our approach was to empower the adolescent participants so that they could effectively manage their own sleep health by engaging them directly in the decision-making process and reinforcing their competencies in adhering to the intervention. The aim here was to promote participants' "buy-in" to the program to maximize their ongoing implementation. We hypothesized two necessary conditions to achieve this aim-participants had to see at least some change due to the intervention and they had to be able to live with the intervention. Therefore, adolescents were explicitly invited to be active members of a "treatment team," in which they were regarded as the experts on their own sleep routine. As such, they were involved in key decision-making processes (eg, they were asked to set their own bedtimes and rise times, and whether they wanted to increase or decrease sleep by 15 or 30 minutes). In a more stringently controlled trial, increasing TIB by 15 minutes weekly might have been the maximum and this might have improved efficacy. However, we viewed this as balancing short-term efficacy with adherence, hypothesizing that improved engagement and adherence would translate to better long-term outcomes. Treatment adherence, results, and feedback at treatment end and follow-up consultations seem to support our decision. For example, Phobos' mother noted at treatment end that the worst for her involved "having to get up at 6:00 AM daily to make sure he gets up," yet at follow-up, she reported that he now effectively self-managed his own sleep routine, and his scores on the sleep condition indicator had continued to improve.

Several limitations must be considered when interpreting the results of the current study. First, generalizability is limited given the small-scale single-case design. However, this does not undermine the value of the study, and the importance of these initial findings should not be discounted. The advantages of small-n research and single-case studies are recognized and promoted elsewhere.<sup>41,47,49,50</sup> Such designs are particularly relevant to samples in which complex individual presentations are considered, such as in the current study, where averaged results would be less meaningful. Second, issues of reliability and validity are often raised to discredit small-*n* designs. The current study followed procedural recommendations by Cohen et al and others a priori to address these types of concerns.41,47,49,50 Third, these results may not generalize to objective sleep measures (eg, actigraphy, polysomnography) given that only self-reported results were used in this preliminary investigation. Yet, participants' self-reported accounts need not be undervalued either-these two participants indicated they benefitted considerably from the treatment and we suggest that their engagement in conjunction with our focus on "buy-in" affected this. Nonetheless, where economically viable, objective sleep measures are recommended. Fourth, although we advocated for evaluation of potential adverse effects associated with this brief intervention, admittedly we did not assess cognitive functioning, academic performance, or other important social and individual risk-taking behaviors. There may be risks and adverse effects associated with this intervention that we did not consider, identify, or discover through follow-up interviews. Nonetheless, we considered this study a necessary first step before engaging in a costly and potentially dangerous larger scale trial, which we suggest is now warranted. In addition, with respect to TST, the current intervention was designed to prioritize sleep quality over quantity by taking a brief interventions approach and such an approach may be open to criticism. We hypothesized, however, that producing rapid short-term change in sleep quality might translate into longerterm gains in sleep quantity. Although our data did not allow us to fully test this, if the observed trend continued, one would expect to see increased TST with increased TIB if SE remained stable. Although only a snapshot of the week preceding followup, this trend was supported via both participants' improved TST and SE. Future research might evaluate this further by recording sleep data throughout the intervening period, and perhaps preceding different follow-up schedules (eg, at 6 and 12 months).

To conclude, the aim of this brief 3-week behavioral intervention for adolescent insomnia was to improve sleep outcomes, and in this regard, the current study was successful. It appeared that through empowering adolescents to change their sleep-wake cycles and experience better sleep outcomes in the short term, these same adolescents appeared motivated and capable of engaging in their own effective and self-managed treatment and they reported experiencing better sleep outcomes in the longer-term. These findings are encouraging in that they support larger-scale clinical evaluation of a brief, safe, and effective intervention that has the potential to effect rapid positive change in adolescent sleep.

#### ABBREVIATIONS

BBT-I, brief behavioral treatment for insomnia CBT, cognitive behavioral therapy CBT-I, cognitive behavioral therapy for insomnia DASS-Y, Depression, Anxiety, Stress Scale for Youth FFS, Flinders Fatigue Scale CBQ-20, Conflict Behavior Questionnaire (20-item short form) RCI, reliable change index SCI, Sleep Condition Indicator SE, sleep efficiency SEE, sleep efficiency enhancement SOL, sleep onset latency SRT, sleep restriction therapy TIB, time in bed TST, total sleep time

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# DISCLOSURE STATEMENT

All authors have seen and approved this manuscript for publication. The authors report no conflicts of interest.