The Journal of Child Psychology and Psychiatry

Journal of Child Psychology and Psychiatry 63:11 (2022), pp 1423–1433 doi:10.1111/jcpp.13590

Sleeping Sound Autism Spectrum Disorder (ASD): a randomised controlled trial of a brief behavioural sleep intervention in primary school-aged autistic children

Nicole Papadopoulos,[1,2] Emma Sciberras,[1,3,4] Harriet Hiscock,[3,4,5] Katrina Williams,[3,4,6] Jane McGillivray,[7] Cathrine Mihalopoulos,[8] Lidia Engel,[8] Matthew Fuller-Tyszkiewicz,[7] Susannah T. Bellows,[1] Deborah Marks,[5] Patricia Howlin,[9] and Nicole Rinehart[1,2]

[1]Deakin Child Study Centre, Deakin University, Burwood, VIC, Australia; [2]School of Educational Psychology and Counselling, Faculty of Education, Monash University, Clayton, VIC, Australia; [3]Murdoch Children's Research Institute, Parkville, VIC, Australia; [4]Department of Paediatrics, University of Melbourne, Parkville, VIC, Australia; [5]Royal Children's Hospital, Parkville, VIC, Australia; [6]Department of Paediatrics, Monash Children's Hospital, Monash University, Clayton, VIC, Australia; [7]Deakin University, Geelong, VIC, Australia; [8]Deakin Health Economics, Institute for Health Transformation, Deakin University, Burwood, VIC, Australia; [9]Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, UK

Background: Behavioural sleep problems are common in children with autism spectrum disorder (ASD); however, evidence for the efficacy of behavioural sleep interventions is limited. This study examined the efficacy of a brief behavioural sleep intervention in autistic children. It was hypothesised that the intervention would reduce overall child sleep problems (primary outcome), in addition to improvements in children's social, emotional, cognitive, academic functioning, and quality of life, and parent/caregivers' stress, quality of life, and mental health (secondary outcomes). **Methods:** A randomised controlled trial was conducted with participants randomised via a computer-generated sequence to the sleeping sound intervention (n = 123) or treatment as usual (n = 122) group. Participants comprised 245 children with an ASD diagnosis. Inclusion criteria were as follows: confirmation of DSM IV or DSM-5 diagnosis of ASD, participants aged between 5 and 13 years and parent/caregiver report of moderate–severe sleep problems. Exclusion criteria were as follows: parent/caregiver intellectual disability or lacking sufficient English to complete questionnaires; and child participant with co-occurring medical conditions known to impact sleep. The intervention group received the sleeping sound intervention (2 × 50-min face-to-face sessions plus follow-up phone call) by a trained clinician. **Results:** Change in children's sleep problems was measured by the Children's Sleep Habits Questionnaire (CSHQ) at 3 months post randomisation. Parents/caregivers of children in the intervention group reported a reduction in child sleep problems at 3 months post randomisation (effect size: E.S −0.7). There were also small effects in a number of child (internalising symptoms, emotional behavioural disturbance and quality of life) and parent/caregiver (mental health, parenting stress and quality of life) outcomes; however, these did not remain significant when controlling for multiple comparisons. **Conclusions:** The sleeping sound ASD intervention is an efficacious and practical way to reduce sleep problems for autistic children. This brief behavioural intervention has the potential to be embedded easily into the Australian healthcare system. **Keywords:** Autism spectrum disorders; sleep; treatment trial; RCT design; intervention.

Introduction

Autism spectrum disorder (hereafter 'autism'[1]) is estimated to affect at least 1% of people worldwide (Fombonne, 2018). Core symptoms include social communication disturbance and restricted and repetitive behaviours (American Psychiatric Association, 2013). Autism is also associated with many other psychological (e.g., anxiety disorder and attention deficit/hyperactivity disorder [ADHD]), neurological, and medical conditions (Matson, Matson, & Beighley, 2011; Simonoff et al., 2008). Approximately 40–80% of children with autism experience behavioural sleep difficulties (Malow & McGrew, 2008; Souders et al., 2009), the most common being difficulties with sleep onset (e.g., settling and falling asleep), sleep maintenance (e.g., frequent night wakings), and reduced sleep duration (Richdale & Schreck, 2009). Although the aetiology of sleep problems is unclear, biopsychosocial factors are indicated (Richdale & Schreck, 2009).

While pharmacological interventions such as melatonin are commonly prescribed to assist families in reducing sleep problems, the National Institute for Health and Care Excellence (NICE) guidelines recommend behaviourally based sleep interventions as a first-line treatment (NICE, 2013). Nevertheless, empirical evidence for the efficacy of behavioural sleep interventions in autistic children is limited. Systematic reviews highlight inconclusive findings,

Conflict of interest statement: No conflicts declared.

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.
This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

attributed to methodological heterogeneity across studies (e.g., differences in length of intervention, outcome measures, and assessment of sleep problems), lack of rigorous randomised controlled trials (RCTs), and inconsistency in the evaluation of secondary child and family outcomes (Beresford et al., 2018; Pattison, Papadopoulos, Marks, McGillivray, & Rinehart, 2020; Rigney et al., 2018; Scantlebury et al., 2018).

Three randomised controlled trials have explored behavioural interventions for sleep problems in children with autism. Adkins et al., (2012) found no effect on children's sleep problems using a brief psychoeducational intervention (n = 18) delivered via a pamphlet on insomnia compared to a control group (n = 18) 2 weeks postintervention. Johnson et al. (2013), using an individualised behavioural sleep programme (n = 15), found significant improvements in sleep problems with small-to-medium effects compared to a comparison group (n = 18) receiving psychoeducational material over an 8-week period. Malow et al. (2014) reported large and significant effects in improving sleep problems as well as improvements in child (behavioural functioning and quality of life) and parenting (sense of competence) outcomes for both an individualised (n = 41) and group-based (n = 39) behavioural sleep intervention 1-month postintervention, although this study did not include a control group. These mixed findings indicate the need for larger and more rigorous studies.

In 2018, we conducted a pilot study that involved reanalysis of our sleeping sound with ADHD trial data (Hiscock et al., 2015) of a brief behavioural sleep intervention in a sample of 61 children aged 5–13 years with a co-occurring diagnosis of autism and ADHD (Papadopoulos, Sciberras, Hiscock, Mulraney, et al., 2019). The purpose of the reanalysis was to examine whether the intervention was also effective in treating sleep problems in a subgroup of children with co-occurring ADHD and ASD. The sleeping sound intervention (Hiscock et al., 2015), initially designed for typically developing children (Quach, Hiscock, Okoumunne, & Wake, 2011), tailors behavioural sleep strategies to the family over two face-to-face sessions and a follow-up phone call with a clinician. Children (n = 28) who received the sleeping sound intervention showed sleep improvements at 3 (moderate-to-large effect size; −0.7) and 6 (moderate effect size; −0.5) months postrandomisation compared with treatment as usual (TAU) controls (n = 33). Families in the intervention group also reported improvements in psychosocial functioning at 3 and 6 months postrandomisation with small-to-moderate effect sizes reported across all psychosocial measures.

The present study aimed to investigate the efficacy of the sleeping sound intervention in reducing sleep problems in a RCT of children with a primary diagnosis of autism (aged 5–13 years) compared with TAU controls receiving standard clinical or community care. Secondary aims included investigating the impact of the intervention on children's social, emotional, cognitive and academic functioning, and quality of life, and parent/caregivers' stress, quality of life, and mental health. We hypothesised that children receiving the sleeping sound intervention would show reduced overall sleep problems at 3 months (primary outcome), as well as improvements in secondary outcomes measuring child and parent/caregiver outcomes (see Table 1), compared with the TAU group.

Methods

The study protocol is published (Papadopoulos, Sciberras, Hiscock, Williams, et al., 2019) and the trial is registered with the International Trial Registry (ISRCTN14077107). Human Research Ethics Committees from the Royal Children's Hospital Melbourne (36154), Deakin University (2017-130), Victorian Department of Education and Early Childhood Development (2016_003134) and the Catholic Education Office Melbourne (0501) approved this study.

Study design and criteria

This RCT was conducted at Deakin University and The Royal Children's Hospital, Melbourne, Australia. Children aged 5 through 13 years were recruited from referrals from Victorian paediatric clinics (50%, n = 122) and by advertising in clinical, research, and community networks (50%, n = 123). Inclusion criteria were as follows: (i) written evidence of a clinically confirmed, DSM IV or DSM-5 multidisciplinary diagnosis of ASD or confirmation by the treating paediatrician; (ii) clinical cut-off score ≥ 11 for ASD symptom severity on the Social Communication Questionnaire–Lifetime form (Rutter, Bailey, & Lord, 2003); (iii) a parent/caregiver-reported moderate-to-severe behavioural sleep problem persisting for ≥ 4 weeks; and (iv) at least one parent/caregiver-reported child sleep problem for chronic insomnia and/or delayed sleep–wake phase as defined by the International Classification of Sleep Disorders–Third-Edition diagnostic criteria (American Academy of Sleep Medicine, 2014).

Exclusion criteria were as follows: parent/caregiver-reported child intellectual disability, co-occurring medical condition known to impair sleep (e.g., epilepsy, blindness, traumatic brain injury, and neuropsychiatric disorders such as Tourette's syndrome), genetic conditions related to intellectual impairment (e.g., Down Syndrome, Tuberous Sclerosis, Fragile X, and rare genetic abnormalities affecting brain development), or suspected obstructive sleep apnoea (OSA). Children presenting with other chronic conditions that had the potential to affect sleep were screened by a study paediatrician and excluded if appropriate. Parents/caregivers of children with suspected OSA (as indicated by parent endorsement of three Sleep Disordered Breathing items on the Children's Sleep Habits Questionnaire) were asked to complete further assessment with a study paediatrician over the phone to ascertain eligibility. In a clinical interview, children with current symptoms of OSA such as snoring, apnoea, daytime tiredness, and a history of recurrent upper airways disease were excluded from the study and referred to their community paediatrician for further clinical investigation and treatment. After approximately 6 months, families were followed up by a study paediatrician to ascertain if they were eligible to participate (i.e., if OSA symptoms have resolved and the child continues to have ongoing sleep problems). Parents/caregivers lacking sufficient English to complete the study questionnaires were

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

Table 1 Study outcome measures, measured at baseline, 3 months, and 6 months

Primary Outcome: Child
Overall child sleep problems
Children's Sleep Habits Questionnaire (CSHQ). Thirty-three-item parent report validated measure of sleep that can distinguish clinical from community samples. Provides a measure of total sleep problems and eight subscale scores reflecting major behavioural sleep disorders (bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night waking, parasomnias, sleep disordered breathing, and daytime sleepiness).
Secondary Outcomes: Child
Sleep Hygiene
Sleep Hygiene Scale. Six-item study developed parent report measure adapted from the Bedroom Routines Scale (see Sciberras, Song, Mulraney, Schuster, & Hiscock, 2017) and considers existence of set routines before bed and regular bed time, where the child falls asleep, and whether potentially distracting or stimulating items are present in the child's bedroom. Each item is dichotomised and scored to give a composite score. Higher scores indicate poorer sleep hygiene.
Daytime Sleepiness
Teacher Daytime Sleepiness Questionnaire. 10-item validated teacher report scale of daytime sleepiness at school.
Emotional and behavioural problems
Developmental Behavioural Checklist (DBC).[a] Ninety-six-item parent report measure of emotional and behavioural disturbance in children. Provides a rating of overall behavioural disturbance and five subscales: disruptive/antisocial behaviour, self-absorbed, communication disturbance, anxiety, and ASD social relating.
Strengths and Difficulties Questionnaire (SDQ), parent and teacher versions. Twenty-five items assessing the following subscales: hyperactivity/inattention, conduct problems, emotional symptoms, peer relationship problems, and prosocial behaviour.
Social communicative symptoms
Social Communication Questionnaire (SCQ) Current. Forty-item parent report measure of ASD symptoms in past 3 months to measure change in ASD social communication symptoms over time.
Cognitive performance[b]
NIH Toolbox. Cognitive Domain Tasks to assess cognitive functioning. iPad administered assessment of executive function, attention, episodic/working memory, processing speed, language abilities, new learning, and reading.
Academic achievement[b]
Two subtests from the Wide Range Achievement Test (WRAT) assessing spelling and math computation skills.
School attendance
Parent report of whether or not their child had missed school over the preceding 3 months, and the number of days missed school during that period.
Quality of life
Child Health Utility 9D (CHU9D). Nine-item parent proxy measure of child quality of life.
Secondary Outcomes: Parent
Stress
Parenting Stress Index 4SF (PSI-4SF). Thirty-six-item measure of parenting stress. Provides a measure of total parenting stress and three subscales reflecting the major sources of parenting stress (parental distress, difficult child, and parent–child dysfunctional interaction).
Mental Health
Kessler 10 (K10). A 10-item validated measure of adult psychological distress.
Quality of Life
Assessment of Quality of Life (AQoL4D). Twelve-item measure of parent quality of life.
Work attendance
Parent report of whether or not they had missed paid work over the preceding 3 months in order to care for their child, and the number of hours of missed from paid work during that period.

[a]Measured only at baseline and 6 months postrandomisation.
[b]Measured only at 6 months postrandomisation.

also excluded. Children taking melatonin and other medications remained in the trial if they met all other inclusion criteria. Participants in the TAU group received standard services and support currently available in the community. There are currently no 'standard' services/interventions available in Australia for children with ASD who have sleep disorders. Web-based 'tip sheets' are available on the topic of ASD sleep management, but these are currently of varying quality and are based on psychoeducation related to promoting healthy sleep practices.

Procedure

Parents/caregivers who registered interest in the study completed a screening questionnaire with a member of the research team via telephone to confirm eligibility. Eligible and interested parents/caregivers were then asked to complete a consent form, provide confirmation of ASD diagnosis from their paediatrician where applicable, and complete a baseline survey. Surveys were completed on REDCap, a secure research database, or hardcopy at baseline and 3 and 6 months postrandomisation. Researchers contacted the child's nominated school teacher to collect teacher-reported data if the parent/caregiver consented (n = 228 parent/caregiver consent for teacher contact; n = 158 teacher data collected) (See CONSORT flow diagram, Figure 1). Cognitive testing was completed at 6 months postrandomisation during a face-to-face assessment with the child. Assessments took approximately 60 min to complete, and took place across several locations in Victoria (The Royal Children's Hospital in Melbourne, Deakin University campuses in Burwood and Geelong, or a home visit if preferred by parents/caregivers).

Measures

Outcome measures are summarised in Table 1. Measures were administered at baseline, 3 and 6 months postrandomisation. The primary outcome measure was children's sleep problems

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

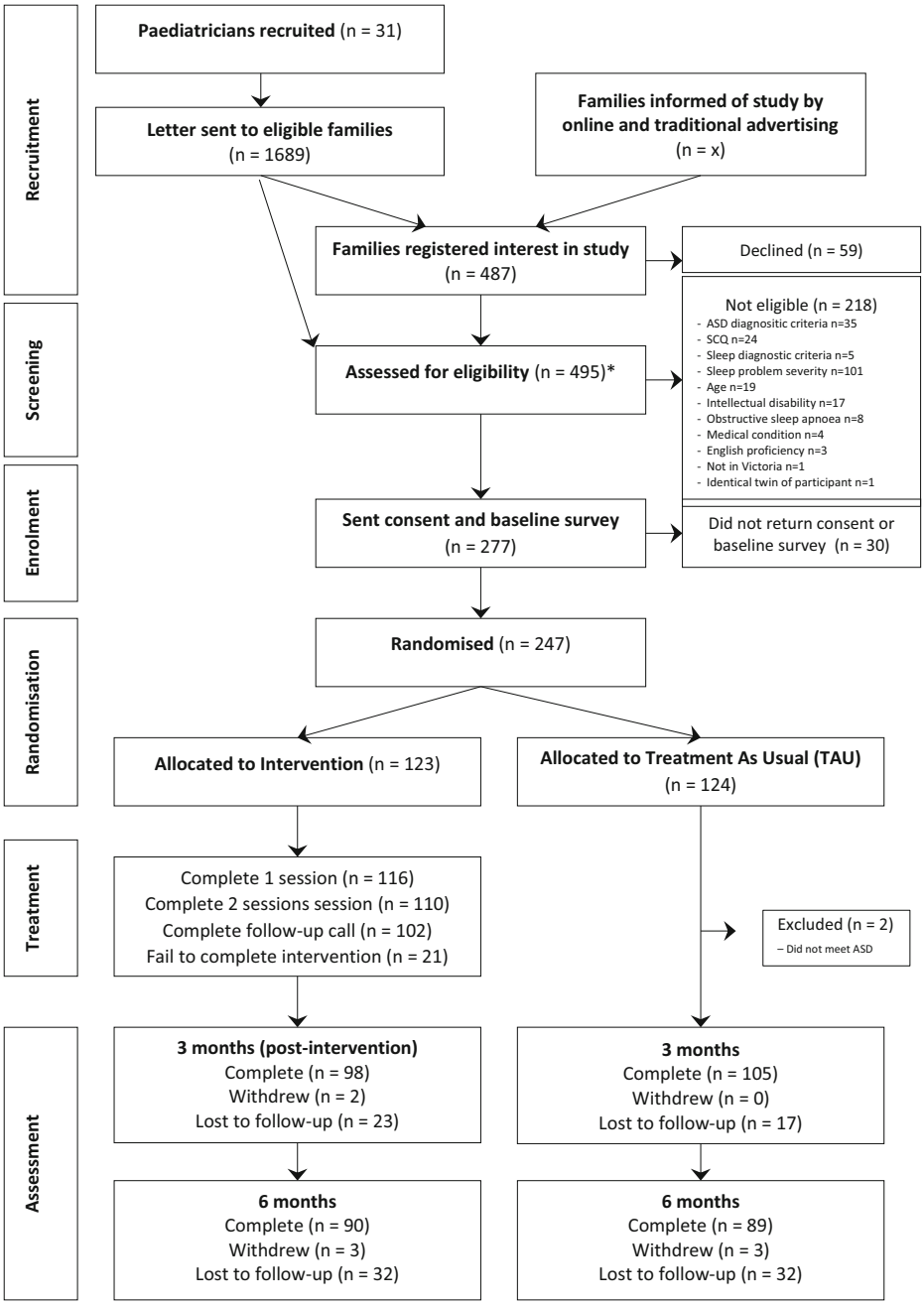

Figure 1 Flow of participants. * Includes 428 participants who registered interest via opt-in recruitment approach, and 67 participants who were contacted via the opt-out recruitment method. See protocol paper Papadopoulos et al. BMJ Open 2019 for further details

as measured by the CSHQ at 3 months postrandomisation. Secondary outcome measures included parent and child quality of life; child social, emotional, cognitive, and academic functioning; child sleep hygiene and daytime sleepiness; child school attendance; parent work attendance; and parent stress and mental health.

Randomisation and masking

After completion of the baseline survey, 247 participants were cluster randomised (according to family unit) to the intervention (INT) group or the TAU control group. Two participants were excluded postrandomisation as they were found to not meet diagnostic inclusion criteria postenrolment. An independent researcher allocated participants using a computer-generated randomisation sequence with 1:1 ratio between groups and blocks of randomly varying size (4, 6, and 8). Randomisation was stratified by parent-reported child gender. Families with siblings enrolled in the study were randomised in sequence based on the return of the primary caregiver baseline surveys, with all subsequent siblings allocated to the same treatment group arm (INT = 10 pairs, TAU = 9 pairs). Selected members of the research team, the statistician, and chief investigators were blinded to group allocation.

Intervention description

The sleeping sound intervention involved children and parents/caregivers attending two consecutive 50-minute face-to-face sessions and a follow-up phone call after the second face-

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

to face session, all at 2-week intervals, delivered by a clinician (e.g., paediatrician or psychologist) experienced in working with children with autism. Prior to delivery of the intervention, all clinicians received training on paediatric sleep management over two 3-hr interactive sessions. To ensure the fidelity of the programme, clinicians followed a standardised manual and met fortnightly to discuss any clinical issues. The first intervention session focused on assessing the type and likely cause of the child's specific sleep difficulties through parent and child report and setting goals in relation to these. Clinicians provided families with psychoeducation about normal sleep patterns, their child's specific sleep problem(s), and developing healthy sleep practices. Parents and children were then invited to choose from a suite of behavioural sleep strategies to develop an individualised sleep management plan that was tailored to the child's needs and the family's capacity for implementation. Clinicians recommended strategies for particular behavioural sleep disturbances, such as bedtime fading for delayed sleep phase and prolonged night waking, graduated extinction (e.g., camping out or parental checking) for children needing parental presence at sleep time, and the use of a 'bedtime pass' for bedtime resistance. Individually tailored strategies were based on interventions used in the previous sleeping sound with ADHD trial (Hiscock et al., 2015). Adaptations to the intervention materials were made to facilitate delivery of the intervention for children with autism. These included the development of a range of visual tools (e.g., visual sleep schedules and social scripts) to reinforce learning. Parents were given written information sheets to support the discussion and sleep diaries to monitor their child's progress. The second session was used to reinforce sleep strategies, monitor sleep patterns (by reviewing a sleep diary), and to address any implementation difficulties reported by parents/caregivers. Parents and children were given the option to try other strategies from the recommended list if they encountered difficulties with implementation. A follow-up phone call 2 weeks later provided further support and an opportunity to reinforce strategies. The face-to-face sessions took approximately 50 min to complete, while the follow-up phone call was completed in approximately 30 min. Compliance with the intervention was assessed by documenting if participants attended all three intervention sessions (See Papadopoulos, Sciberras, Hiscock, Williams, et al., 2019).

Sample size

Required sample size was powered for a 0.5 standardised mean difference at 3 and 6 months postrandomisation for the primary outcome (a more conservative estimate than found in our pilot study), with alpha set at .05 (two tailed), power = .80, and allowances of 20% attrition due to loss of participants to follow-up and corrections in effective sample size attributable to clustering effects of individual and paediatrician (Papadopoulos, Sciberras, Hiscock, Williams, et al., 2019). On this basis, we aimed for 117 participants per group at baseline (234 children in total).

Statistical analysis

Analyses were undertaken using Stata version 16 and conducted on an intention-to-treat basis, with participant data included as per initial treatment group allocation. Linear mixed models were used for continuous outcomes; count-based outcomes (days off work and time missed from school) were modelled using mixed-effects negative binomial models to account for positive skew in these variables. Changes over time in outcome variables were modelled in the same model by regressing outcome scores onto dummy variables reflecting baseline versus 3-month follow-up and baseline versus 6-month follow-up time points. The interaction between these dummy variables and group thus tested group differences in change in variables over time. Although our protocol proposed that the repeated time points of data (Level 1) may exhibit clustering effects by individual (Level 2) and also by family and/or treating paediatrician (Level 3), in most instances the random effects for family unit and paediatrician did not significantly deviate from 0 based on comparison of log likelihood values for models with and without random intercepts at Level 3. Thus, with the exception of outcomes that had a significant random intercept for family unit or paediatrician, outcomes were tested with two-level mixed models, where data at each time point were clustered by individual for within-participant effects.

Models are reported in unadjusted form, as well as adjusting for covariates identified a priori: parent-reported child gender, age, ASD symptom severity, medication use, and socioeconomic status (SES). Analyses for primary and secondary outcomes were rerun per protocol, excluding intervention participants who failed to complete all treatment modules. Consistent with our protocol, primary outcomes were tested with unadjusted p values, whereas p values reported for secondary outcomes were corrected for risk of Type I error inflation using the Benjamini–Hochberg approach (Benjamini & Hochberg, 1995).

In these models, missing data were handled using conditional maximum likelihood estimation. However, as this approach makes an untestable assumption that missingness is ignorable, sensitivity analyses were conducted to evaluate robustness of attained results to possible presence of non-ignorable patterns of missingness (i.e., not missing at random; NMAR). Pattern mixture models via the Mimix package (Cro, Morris, Kenward, & Carpenter, 2016) were used for sensitivity analysis. Several plausible NMAR patterns were tested with Mimix: (a) last mean carried forward, which imputes the mean at the previous time point from one's assigned group; (b) jump to reference, in which an individual's missing data are imputed with the mean value from the control group at that time point; and (c) copy increments in reference, in which an individual's missing data are imputed with the mean increment from the previous time point for the control group regardless of treatment assignment at baseline. Interim missingness (i.e., when a participant misses a time point but returns for a later wave) was treated as MAR, which is a reasonable assumption for intermittent response rather than complete dropout (Cro et al., 2016). Fifty imputations were undertaken per model.

Results

Baseline characteristics

Table 2 describes baseline characteristics postrandomisation for the two groups. Overall, most children were male; were using prescription sleep medication (e.g., melatonin); and most primary caregivers identified as female. Many participants had at least one parent/caregiver-reported co-occurring diagnosis. Between-group differences were nonsignificant, except that participants in the intervention group were younger ($t = 2.73$, $p = .01$).

Primary outcomes: Child sleep problems

As shown in Table 3, at 3 months postrandomisation, the unadjusted and adjusted mean group differences in severity of child sleep problems were significant for the majority of the CSHQ subscales with most p values ≤.001 and E.S moderate to large.

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

Table 2 Sample characteristics of participants. Values are numbers (percentages) unless stated otherwise

Characteristic	Intervention group (n = 123)	Control group (n = 122)	Comparison
Children			
Age in years, *M (SD)*, range	8.45 (2.10), 5.09-13.01	9.18 (2.10), 5.28-13.18	t = 2.73, p = .003
Parent-reported gender (F:M)	43 (35%):80 (65%)	41 (34%):81 (66%)	$\chi^2(1)$ = 0.05, p = .823
ASD symptom severity, *M (SD)*, range (SCQ–Lifetime Score)	14.11 (5.82), 3-29 20.27 (5.83), 11-34	14.65 (5.69), 3-29 20.01 (5.12), 11-33	t = 0.73, p = .233 t = −0.36, p = .642
Prescription medication use[b]			
Sleep medication	66 (57%)	51 (47%)	$\chi^2(1)$ = 2.10, p = .147
Stimulant medication	11 (10%)	18 (15%)	$\chi^2(1)$ = 1.48, p = .223
Parent-reported comorbidities:			
Anxiety disorder	53 (43%)	63 (52%)	$\chi^2(1)$ = 1.80, p = .180
Attention deficit/hyperactivity disorder	44 (36%)	52 (43%)	$\chi^2(1)$ = 1.21, p = .272
Oppositional defiant disorder	10 (8%)	14 (11%)	$\chi^2(1)$ = 0.78, p = .378
Depressive disorder	2 (2%)	2 (2%)	$\chi^2(1)$ = 0.00, p = .993
Primary Caregiver			
Age in years, *M (SD)*, range	41.02 (4.80), 29.32, 54.08	41.87 (5.52), 26.15-52.42	t = 1.28, p = .101
Female	119 (97%)	115 (94%)	$\chi^2(1)$ = 0.88, p = .348
Education			$\chi^2(1)$ = 0.85, p = .653
Did not complete high school	11 (9%)	15 (12%)	
Completed high school only	29 (24%)	30 (25%)	
Completed tertiary study	83 (67%)	77 (63%)	
Single parent household	23 (19%)	34 (28%)	$\chi^2(1)$ = 2.88, p = .089
Family socioeconomic disadvantage[a], *M (SD)*, range	1029.80 (55.08), 795-1116	1036.83 (54.85), 795-1117	t = 1.00, p = .159

[a]Participants only reported prescription medication use.
[b]SEIFA (postcode) data.

Effect sizes for CSHQ subscale scores were as follows: bedtime resistance (E.S −0.50), sleep onset delay (E.S −0.87), sleep duration (E.S −0.55), and total CSHQ score (E.S −0.70). Smaller, but still significant effects were found for sleep anxiety (E.S −0.41), night waking (E.S −0.32) and parasomnias (E.S −0.48).

All effects remained significant at 6 months postrandomisation, with the exception of daytime sleepiness (p = .69). These effects remained significant after adjusting for covariates, with the exception of daytime sleepiness at 3 months (p = .09). Furthermore, results that were significant in adjusted analyses tended to remain significant in sensitivity analyses, with two exceptions: (a) the significant effect of sleep onset delay at 6 months was nonsignificant in one of the three NMAR analyses (p = .07); and (b) the significant effect of parasomnias at 6 months was nonsignificant in one of the three NMAR analyses (p = .08). Thus, conclusions based on significance tests remain largely consistent following adjustment for covariates and possibility of plausible nonignorable missing data patterns.

Secondary outcomes

Child outcomes. Table S1 shows effects of treatment on secondary outcomes, with Table S2 providing subscale scores. In the adjusted analysis, none of the effects were significant. This is not only a function of correcting for multiple comparisons but also small effect sizes. When we did not adjust for multiple comparisons, the following child secondary outcomes were significant at 3 months (SDQ subscale: emotional problems and child quality of life) and 6 months (DBC total; DBC subscales; disruptive/antisocial behaviours; self-absorbed; and anxiety and SDQ: emotional problems) postrandomisation. Significant parent/caregiver outcomes at 3 months postrandomisation included: psychological distress (K10), parental distress (PSI total score); PSI subscales: parenting distress (PD); difficult child (DC); parent–child dysfunctional

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

Table 3 Time × Group interaction effects for CSHQ

	Control group		Intervention group		Unadjusted difference (intervention–control)			Adjusted difference (intervention–control)		
Outcomes	*n*	*M* (*SD*)	*n*	*M* (*SD*)	*M* (95% *CI*)	Effect size	*p*	*M* (95% *CI*)	Effect size	*p*
CSHQ Total										
Baseline	122	57.79 (9.20)	123	56.32 (8.71)						
3 months	105	56.04 (9.05)	97	47.74 (8.23)	−6.27 [−7.93, −4.61]	−0.70	<.001	−6.00 [−7.76, −4.23]	−0.67	<.001
6 months	88	55.12 (9.13)	90	48.74 (8.10)	−4.45 [−6.22, −2.67]	−0.50	<.001	−3.81 [−5.70, −1.91]	−0.43	<.001
CSHQ Bedtime Resistance Subscale										
Baseline	122	10.03 (3.19)	123	10.80 (3.32)						
3 months	105	9.65 (3.24)	97	8.48 (2.80)	−1.63 [−2.25, −1.02]	−0.50	<.001	−1.65 [−2.31, −0.99]	−0.51	<.001
6 months	88	9.46 (2.98)	90	8.67 (2.64)	−1.16 [−1.71, −0.60]	−0.36	<.001	−1.13 [−1.73, 0.53]	−0.35	<.001
CSHQ Sleep Onset Delay Subscale										
Baseline	122	2.45 (0.71)	122	2.48 (0.67)						
3 months	105	2.26 (0.77)	96	1.67 (0.66)	−0.60 [−0.81, −0.40]	−0.87	<.001	−.59 [−0.81, −0.38]	−0.85	<.001
6 months	88	2.18 (0.74)	90	1.88 (0.83)	−0.30 [−0.53, −0.07]	−0.43	.011	−0.26 [−0.51, −0.01]	−0.38	.04
CSHQ Sleep Duration Subscale										
Baseline	122	6.12 (1.64)	123	6.15 (1.68)						
3 months	105	5.65 (1.64)	97	4.72 (1.52)	−0.91 [−1.36, −0.46]	−0.55	<.001	−0.86 [−1.33, −0.39]	−0.52	<.001
6 months	88	5.74 (1.70)	90	4.84 (1.53)	−0.93 [−1.34, −0.53]	−0.56	<.001	−0.93 [−1.37, −0.50]	−0.56	<.001
CSHQ Sleep Anxiety Subscale										
Baseline	122	7.70 (2.53)	123	7.84 (2.54)						
3 months	105	7.60 (2.26)	97	6.54 (2.37)	−1.05 [−1.53, −0.56]	−0.41	<.001	−1.00 [−1.52, −0.47]	−0.39	<.001
6 months	88	7.56 (2.33)	90	6.68 (2.24)	−0.79 [−1.32, −0.26]	−0.31	.003	−0.76 [−1.32, −0.20]	−0.30	.01
CSHQ Night Waking Subscale										
Baseline	122	5.37 (1.94)	123	5.53 (1.88)						
3 months	105	5.12 (1.91)	97	4.62 (1.60)	−0.61 [−0.95, −0.26]	−0.32	.001	−0.67 [−1.04, −0.30]	−0.35	<.001
6 months	88	5.10 (1.75)	90	4.70 (1.68)	−0.54 [−0.94, −0.14]	−0.28	.009	−0.58 [−1.01, −0.16]	−0.30	.01
CSHQ Parasomnias Subscale										
Baseline	122	10.73 (2.52)	123	10.32 (2.15)						
3 months	105	10.90 (2.38)	97	9.31 (1.95)	−1.13 [−1.63, −0.63]	−0.48	<.001	−0.99 [−1.50, −0.48]	−0.42	<.001
6 months	88	10.67 (2.40)	90	9.34 (2.21)	−0.95 [−1.51, −0.40]	−0.41	.001	−0.76 [−1.35, −0.17]	−0.32	.01
CSHQ Sleep Disordered Breathing Subscale										
Baseline	122	3.64 (1.06)	123	3.48 (0.86)						
3 months	105	3.78 (1.11)	97	3.41 (0.81)	−0.20 [−0.44, 0.04]	−0.21	.096	−0.20 [−0.46, 0.06]	−0.21	.14
6 months	88	3.77 (1.16)	90	3.46 (1.07)	−0.14 [−0.40, 0.13]	−0.15	.320	−0.12 [−0.41, 0.17]	−0.12	.43
CSHQ Daytime Sleepiness Subscale										
Baseline	122	15.24 (3.53)	123	13.61 (3.39)						
3 months	105	14.57 (3.51)	97	12.13 (3.41)	−0.85 [−1.61, −0.10]	−0.25	.027	−0.70 [−1.51, 0.11]	−0.20	.09
6 months	88	14.10 (3.43)	90	12.43 (3.31)	−0.16 [−0.97, 0.64]	−0.05	.693	0.26 [−0.57, 1.09]	0.08	.54

CSHQ, Children's Sleep Habits Questionnaire.
p values reflect time x group effects with baseline data compared against 3- and 6-month follow-ups respectively.

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

interaction (PCDI); and difficult child (DC) subscale of the PSI at 6 months postrandomisation. It should be noted that effect sizes reported for the secondary outcomes mentioned above were typically small (as defined by a Cohens $d > 0.2$) (Please see Tables S3 and S4 for secondary outcome results not adjusted for Type 1 error inflation).

Per-protocol analyses

Analyses for primary and secondary outcomes were rerun, excluding individuals in the intervention group who failed to complete all components of the treatment. As compliance levels were high, this resulted in 102 (83%) individuals from the intervention arm retained for per-protocol analyses.

Some results that were significant for the sample overall were nonsignificant for the reduced sample: (a) intervention effects at 3 months were nonsignificant for parent/caregiver-rated SDQ internalising subscale (unadjusted model), K10 (adjusted and unadjusted model), PSI PD subscale (unadjusted and adjusted models), and parent/caregiver's quality of life (unadjusted model); and (b) intervention effects at 6 months were nonsignificant for CSHQ onset delay (covariate adjusted model) (See Table S2 for full set of results for per-protocol analyses).

Discussion

This brief behavioural sleep intervention, tailored to the child's needs, effectively reduced sleep problems at 3 and 6 months postrandomisation in a sample of primary school-aged autistic children. We also found small effects in improvements in several child (internalising symptoms, emotional behavioural disturbance, and quality of life) and parent/caregiver (mental health, parenting stress, and quality of life) outcomes, although when adjusting for multiple comparisons these secondary effects did not remain significant.

The large effect sizes related to improved sleep problems in this study are comparable to the results of previous trials of behavioural and melatonin-based treatments for sleep problems in autism (Malow et al., 2014; Papadopoulos, Sciberras, Hiscock, Mulraney, et al., 2019). Consistent with our previous work evaluating sleeping sound (Papadopoulos, Sciberras, Hiscock, Williams, et al., 2019), we observed a reduction in effect sizes at 6 months postrandomisation. This drop off in effect warrants consideration of a booster session at 6 months follow-up to reinforce behavioural strategies and maintain initial treatment gains.

Our study was not designed to compare melatonin to a behavioural intervention or to assess whether there is a combination effect of the two interventions. As such, evidence for the optimal sequence or combination of melatonin and behavioural intervention is not available. However, children were not excluded from our study if they were taking melatonin. As such, if melatonin therapy is used as first-line therapy, sleep improvements should be monitored, and behavioural therapy advised if sleep problems persist. Ideally, behavioural therapies should be offered first or as an adjunct therapy from the commencement of melatonin. We adjusted a priori based on variables that may hold prognostic value, but these had negligible impact in the current study, including ASD symptom severity. The lack of effect of adjustment for ASD symptom severity in particular is noteworthy, and may suggest that ASD severity does not have a strong influence on change in sleep behaviours over time. Furthermore, our results indicated that children who participated in our study already had good sleep hygiene prior to taking part in the intervention, emphasising the need for behavioural sleep interventions to address sleep problems when healthy sleep habits have already been implemented by families.

We found small effects for a number of secondary child and parent outcome measures for children who received the sleeping sound intervention. These findings are consistent with previous sleep interventions (both behavioural and pharmacological) that show improvement in child and parent functioning (Malow et al., 2012; Reed et al., 2009). Given our results did not remain significant when adjusting for multiple comparisons, further research in larger adequately powered samples is needed to confirm the stability of these findings and to better understand the exact mechanisms that may underlie the differential impact of behavioural sleep interventions on child and parent outcomes for autistic children.

Our study has several strengths. It is the largest RCT examining a brief behavioural sleep treatment in primary school-aged children with autism with moderate-to-severe sleep problems. Participants were from a community sample and, thus, unlike some other studies, we did not exclude children with common co-occurring conditions (e.g., ADHD) or those taking commonly prescribed psychotropic medications, including melatonin and stimulants. Clinically, the sleeping sound protocol is easy to learn and to deliver. The clinicians delivering the intervention had general skills in using behavioural strategies, but no experience in sleep interventions prior to the 6-hour training provided. Engagement of both caregivers and children fostered persistence with the programme and the involvement of children in the selection and implementation of sleep strategies helped mitigate the natural tendency of autistic children to resist change. Finally, the relatively short time frame of the protocol (4 weeks from first session to follow-up phone call) had the advantage of requiring less commitment from families to take part in the programme.

Study limitations include the lack of an objective sleep measure (e.g., actigraphy), although the CSHQ has been found to be highly correlated with objective

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

measures (Souders et al., 2009). Reliance on an unblinded, parent report-based primary outcome measure may also have biased the results. Our findings on child cognitive functioning and academic achievement may be limited by the small number of participants who completed a face-to-face assessment at 6-month follow-up. This was largely attributed to participants being uncontactable to arrange an appointment time, or declining to participate due to reasons such as unavailability. The different assessment settings (for cognitive testing) and different ways in which questionnaire data were captured (online vs. in person or hardcopy) may have also impacted our results. The short time frame of the intervention also meant any family disruptions had a greater impact on implementation of the intervention. Lastly, only a small number of families were able to take part in this study, emphasising the need for the development of ASD interventions that enable increased reach and accessibility for ASD children and families.

Future research investigating the administration of the sleeping sound intervention online (via telehealth) will improve accessibility of the intervention to all families, especially those living in rural and remote communities where access to treatment is limited (Antezana, Scarpa, Valdespino, Albright, & Richey, 2017), and in times of pandemics where families with autistic children may experience an exacerbation of sleep problems (Becker & Gregory, 2020; Panda et al., 2021). Telehealth intervention delivery has grown exponentially in Australia as a result of the current pandemic (Taylor et al., 2021) and holds promise for research and care in the autistic community longer term (Ameis, Lai, Mulsant, & Szatmari, 2020), with research providing preliminary support for this mode of intervention delivery (Ellison, Guidry, Picou, Adenuga, & Davis, 2021). Large-scale translation studies of the implementation of the sleeping sound programme for autistic children by health professionals are also needed. Follow-up on this sample will also allow for an investigation of the long-term benefits of the intervention, including potential mediators and moderators of treatment outcomes. Lastly, a planned health economic evaluation will make it possible to determine its cost effectiveness.

It is increasingly recognised that behavioural sleep problems are highly prevalent in autism and are associated with poor child and family functioning. Brief tailored behavioural sleep interventions such as the sleeping sound intervention offer families safe, nonstigmatised assistance in the treatment of sleep problems, which can easily be embedded in the Australian healthcare system.

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article:

Table S1. Secondary outcomes.
Table S2. Secondary subscale scores.
Table S3. Secondary outcomes not adjusted for multiple comparisons.
Table S4. Secondary outcome subscale scores not adjusted for multiple comparisons.

Acknowledgements

The authors thank the children and their families, paediatricians, and teachers for taking part in this study and the following project staff for their contribution in data collection, supervision, or delivery of the intervention: Amanda Dudley, Ebony Lindor, Christina Martin, Lucy Sommers, Emily Pattison, and Tayla Chellew. The authors have declared that they have no competing or potential conflicts of interest. Open access publishing facilitated by Deakin University, as part of the Wiley - Deakin University agreement via the Council of Australian University Librarians.

Correspondence

Nicole Rinehart, School of Educational Psychology and Counselling, Faculty of Education, Monash University, 19 Ancora Imparo Way, Clayton VIC 3800, Australia; Email: nicole.rinehart@monash.edu

Key points

- Behavioural sleep problems are common in children with autism, and behaviourally based sleep interventions are recommended as a first-line treatment. However, evidence for the efficacy of behavioural sleep interventions is limited.
- This large RCT (n = 245) found significant reductions in parent/caregiver-reported child sleep problems in the intervention group at 3 months postrandomisation (effect size: −0.7). Additional improvements in child outcomes (internalising symptoms, emotional behavioural disturbance, and quality of life) and parent/caregiver outcomes (mental health, parenting stress, and quality of life) were found; however, these did not remain significant when controlling for multiple comparisons.
- This brief behavioural intervention is an efficacious and practical way to reduce sleep problems, with the potential to be embedded easily into the Australian healthcare system.

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

Note

[1] Throughout this document "autism" is used as a shorthand term for Autism Spectrum Disorder. To reflect differing views on terminology among autistic people and their families (Kenny et al., 2016), we use both person first ("child/ren with autism") and identity first (autistic child/") throughout this document.

References

Adkins, K.W., Molloy, C., Weiss, S.K., Reynolds, A., Goldman, S.E., Burnette, C., … & Malow, B.A. (2012). Effects of a standardized pamphlet on insomnia in children with autism spectrum disorders. *Pediatrics*, *130*(Supplement 2), S139–S144.

Ameis, S.H., Lai, M.C., Mulsant, B.H., & Szatmari, P. (2020). Coping, fostering resilience, and driving care innovation for autistic people and their families during the COVID-19 pandemic and beyond. *Molecular Autism*, *11*, 61.

American Academy of Sleep Medicine. (2014). *International Classification of Sleep Disorders (ISCD-3)* (3rd edn). American Academy of Sleep Medicine.

American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders (DSM-5®)*. Washington, DC: American Psychiatric Publishing. Available from: http://ebookcentral.proquest.com/lib/deakin/detail.action?docID=1811753

Antezana, L., Scarpa, A., Valdespino, A., Albright, J., & Richey, J.A. (2017). Rural trends in diagnosis and services for autism spectrum disorder. *Frontiers in Psychology*, *8*, 590.

Becker, S.P., & Gregory, A.M. (2020). Editorial Perspective: Perils and promise for child and adolescent sleep and associated psychopathology during the COVID-19 pandemic. *Journal of Child Psychology and Psychiatry*, *61*, 757–759.

Benjamini, Y., & Hochberg, Y. (1995). Controlling the false discovery rate: A practical and powerful approach to multiple testing. *Journal of the Royal Statistical Society: Series B (Methodological)*, *57*, 289–300.

Beresford, B., McDaid, C., Parker, A., Scantlebury, A., Spiers, G., Fairhurst, C., … & Thomas, M. (2018). Pharmacological and non-pharmacological interventions for non-respiratory sleep disturbance in children with neurodisabilities: A systematic review. *Health Technology Assessment*, *22*, 1–296.

Cro, S., Morris, T.P., Kenward, M.G., & Carpenter, J.R. (2016). Reference-based sensitivity analysis via multiple imputation for longitudinal trials with protocol deviation. *Stata Journal*, *2*, 443.

Ellison, K.S., Guidry, J., Picou, P., Adenuga, P., & Davis, T.E., 3rd (2021). Telehealth and autism prior to and in the age of COVID-19: A systematic and critical review of the last decade. *Clinical Child and Family Psychology Review*, *24*, 599–630.

Fombonne, E. (2018). Editorial: The rising prevalence of autism. *Journal of Child Psychology & Psychiatry*, *59*, 717–720.

Hiscock, H., Sciberras, E., Mensah, F., Gerner, B., Efron, D., Khano, S., & Oberklaid, F. (2015). Impact of a behavioural sleep intervention on symptoms and sleep in children with attention deficit hyperactivity disorder, and parental mental health: Randomised controlled trial. *BMJ*, *350*, h68.

Johnson, C.R., Turner, K.S., Foldes, E., Brooks, M.M., Kronk, R., & Wiggs, L. (2013). Behavioral parent training to address sleep disturbances in young children with autism spectrum disorder: A pilot trial. *Sleep Medicine*, *14*, 995–1004.

Kenny, L., Hattersley, C., Molins, B., Buckley, C., Povey, C., & Pellicano, E. (2016). Which terms should be used to describe autism? Perspectives from the UK autism community. *Autism*, *20*(4), 442–462. https://doi.org/10.1177/1362361315588200

Malow, B., Adkins, K.W., McGrew, S.G., Wang, L., Goldman, S.E., Fawkes, D., & Burnette, C. (2012). Melatonin for sleep in children with autism: A controlled trial examining dose, tolerability, and outcomes. *Journal of Autism and Developmental Disorders*, *42*, 1729–1737.

Malow, B.A., Adkins, K.W., Reynolds, A., Weiss, S.K., Loh, A., Fawkes, D., … & Clemons, T. (2014). Parent-based sleep education for children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *44*, 216–228.

Malow, B.A., & McGrew, S.G. (2008). Sleep disturbances and autism. *Sleep Medicine Clinics*, *3*, 479–488.

Matson, M.L., Matson, J.L., & Beighley, J.S. (2011). Comorbidity of physical and motor problems in children with autism. *Research in Developmental Disabilities*, *32*, 2304–2308.

NICE (2013). Autism spectrum disorder in under 19s: Support and management. Available from: https://www.nice.org.uk/guidance/CG170. [last accessed 10 June 2020].

Panda, P.K., Gupta, J., Chowdhury, S.R., Kumar, R., Meena, A.K., Madaan, P., … & Gulati, S. (2021). Psychological and behavioral impact of lockdown and quarantine measures for COVID-19 pandemic on children, adolescents and caregivers: A systematic review and meta-analysis. *Journal of Tropical Pediatrics*, *67*(1), 1–13. https://doi.org/10.1093/tropej/fmaa122

Papadopoulos, N., Sciberras, E., Hiscock, H., Mulraney, M., McGillivray, J., & Rinehart, N. (2019). The efficacy of a brief behavioral sleep intervention in school-aged children with ADHD and comorbid autism spectrum disorder. *Journal of Attention Disorders*, *23*, 341–350.

Papadopoulos, N., Sciberras, E., Hiscock, H., Williams, K., McGillivray, J., Mihalopoulos, C., … & Rinehart, N. (2019). Sleeping Sound with autism spectrum disorder (ASD): Study protocol for an efficacy randomised controlled trial of a tailored brief behavioural sleep intervention for ASD. *British Medical Journal Open*, *9*, e029767.

Pattison, E., Papadopoulos, N., Marks, D., McGillivray, J., & Rinehart, N. (2020). Behavioural treatments for sleep problems in children with autism spectrum disorder: A review of the recent literature. *Current Psychiatry Reports*, *22*, 46.

Quach, J., Hiscock, H., Okoumunne, O.C., & Wake, M. (2011). A brief sleep intervention improves outcomes in the school entry year: A randomized controlled trial. *Pediatrics*, *4*, 692.

Reed, H.E., McGrew, S.G., Artibee, K., Surdkya, K., Goldman, S.E., Frank, K., … & Malow, B.A. (2009). Parent-based sleep education workshops in autism. *Journal of Child Neurology*, *24*, 936–945.

Richdale, A.L., & Schreck, K.A. (2009). Sleep problems in autism spectrum disorders: Prevalence, nature, & possible biopsychosocial aetiologies. *Sleep Medicine Reviews*, *13*, 403–411.

Rigney, G., Ali, N.S., Corkum, P.V., Brown, C.A., Constantin, E., Godbout, R., … & Weiss, S.K. (2018). A systematic review to explore the feasibility of a behavioural sleep intervention for insomnia in children with neurodevelopmental disorders: A transdiagnostic approach. *Sleep Medicine Reviews*, *41*, 244–254.

Rutter, M., Bailey, A., & Lord, C. (2003). *The social communication questionnaire: Manual*. Los Angeles: Western Psychological Services.

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.

Scantlebury, A., Mcdaid, C., Dawson, V., Elphick, H., Fairhurst, C., Hewitt, C., … & Beresford, B. (2018). Non-pharmacological interventions for non-respiratory sleep disturbance in children with neurodisabilities: A systematic review. *Developmental Medicine & Child Neurology*, *60*, 1076–1092.

Sciberras, E., Song, J.C., Mulraney, M., Schuster, T., & Hiscock, H. (2017). Sleep problems in children with attention-deficit hyperactivity disorder: Associations with parenting style and sleep hygiene. *European Child & Adolescent Psychiatry*, *26*, 1129–1139.

Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: Prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child & Adolescent Psychiatry*, *47*, 921–929.

Souders, M.C., Mason, T.B.A., Valladares, O., Bucan, M., Levy, S.E., Mandell, D.S., … & Pinto-Martin, J. (2009). Sleep behaviors and sleep quality in children with autism spectrum disorders. *Sleep*, *32*, 13.

Taylor, A., Caffery, L.J., Gesesew, H.A., King, A., Bassal, A.-R., Ford, K., … & Ward, P.R. (2021). How Australian health care services adapted to telehealth during the COVID-19 pandemic: A survey of telehealth professionals. *Frontiers in Public Health*, *9*, 1–11. https://doi.org/10.3389/fpubh.2021.648009

Accepted for publication: 19 January 2022

© 2022 The Authors. *Journal of Child Psychology and Psychiatry* published by John Wiley & Sons Ltd on behalf of Association for Child and Adolescent Mental Health.