

# BMJ Open Does a brief, behavioural intervention, delivered by paediatricians or psychologists improve sleep problems for children with ADHD? Protocol for a cluster-randomised, translational trial

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## ABSTRACT

**Introduction:** Up to 70% of children with attention-deficit/hyperactivity disorder (ADHD) experience sleep problems. We have demonstrated the efficacy of a brief behavioural intervention for children with ADHD in a large randomised controlled trial (RCT) and now aim to examine whether this intervention is effective in real-life clinical settings when delivered by paediatricians or psychologists. We will also assess the cost-effectiveness of the intervention.

**Methods and analysis:** Children aged 5–12 years with ADHD (n=320) are being recruited for this translational cluster RCT through paediatrician practices in Victoria and Queensland, Australia. Children are eligible if they meet criteria for ADHD, have a moderate/severe sleep problem and meet American Academy of Sleep Medicine criteria for either chronic insomnia disorder or delayed sleep–wake phase disorder; or are experiencing sleep-related anxiety. Clinicians are randomly allocated at the level of the paediatrician to either receive the sleep training or not. The behavioural intervention comprises 2 consultations covering sleep hygiene and standardised behavioural strategies. The primary outcome is change in the proportion of children with moderate/severe sleep problems from moderate/severe to no/mild by parent report at 3 months postintervention. Secondary outcomes include a range of child (eg, sleep severity, ADHD symptoms, quality of life, behaviour, working memory, executive functioning, learning, academic achievement) and primary caregiver (mental health, parenting, work attendance) measures. Analyses will address clustering at the level of the paediatrician using linear mixed effect models adjusting for potential a priori confounding variables.

**Ethics and dissemination:** Ethics approval has been granted. Findings will determine whether the benefits of an efficacy trial can be realised more broadly at the population level and will inform the development of clinical guidelines for managing sleep problems in this population. We will seek to publish in leading international paediatric journals, present at major conferences and through established clinician networks.

## Strengths and limitations of this study

- First translational trial to determine whether a brief, behavioural sleep intervention has benefits for children with attention-deficit/hyperactivity disorder (ADHD) when delivered in 'real-life' clinical settings by paediatricians and psychologists.
- The inclusion of blinded teacher reports, as well as blinded direct assessment measures to minimise bias.
- Inclusion of economic analyses, which provide data on the cost-effectiveness of the programme.
- Limited generalisability to non-English-speaking families and children with ADHD presenting with serious medical conditions and/or intellectual disability.
- Sleep problems assessed using non-blinded parent report as opposed to objective measures.

**Trial registration number:** ISRCTN50834814, Pre-results.

## INTRODUCTION

Sleep problems are common and more persistent in children with attention-deficit/hyperactivity disorder (ADHD), and associated with poorer child and family well-being.<sup>1–4</sup> Efficacy randomised controlled trials (RCTs) have shown that sleep problems are amenable to intervention<sup>5–10</sup> in children with ADHD, particularly behavioural interventions.<sup>8–10</sup> However, it is unknown whether the same benefits will be observed when such interventions are delivered by practitioners in 'real life' clinical settings. This article describes the protocol for the Sleeping Sound with ADHD Translational RCT, which aims to assess the effectiveness of

a behavioural sleep intervention designed for children with ADHD, when delivered by psychologists or paediatricians in their daily clinical practice.

### Sleep problems in children with ADHD

Up to 70% of children with ADHD experience sleep problems,<sup>1 2</sup> compared with 20–30% of children in the general population.<sup>11</sup> Sleep problems experienced by children with ADHD largely comprise difficulties initiating and/or maintaining sleep, with the most common sleep problems being difficulty falling asleep (reported by 84% of parents of children who report a sleep problem for their child with ADHD), bedtime resistance (68%), tiredness on waking (62%) and difficulty waking in the morning (56%).<sup>2</sup> Medical conditions such as obstructive sleep apnoea (OSA) can also affect sleep in children with ADHD, although behavioural sleep problems appear more common.<sup>12</sup> In this study, we use the term sleep problems to denote those difficulties that can be addressed using behavioural intervention. Sleep problems have been associated with worse functioning for children with ADHD, including poorer quality of life (QoL), behaviour, daily functioning and working memory, as well as increased school absenteeism.<sup>2 4 13</sup> They thus represent an important modifiable target for intervention.

A number of factors have been associated with sleep problems in children with ADHD and it is possible that biological sleep problems such as restless legs syndrome, OSA and narcolepsy can lead to continued sleep disruptions.<sup>12</sup> Insomnia can be a side effect of stimulant medication,<sup>1 14</sup> although unmedicated children with ADHD also experience higher rates of sleep problems relative to non-ADHD controls.<sup>15</sup> Comorbid externalising and internalising comorbidities are each associated with a twofold increase in moderate/severe sleep problems in children with ADHD, while co-occurring internalising and externalising comorbidities are associated with a threefold increase.<sup>16</sup> Parenting inconsistency in daily household routines has also been associated with increased bedtime resistance in this group.<sup>17</sup> Biologically, there is overlap in the brain regions and genetic factors (eg, catecholaminergic system, CLOCK genes) associated with ADHD and sleep disorders/arousal regulation.<sup>1</sup> ADHD is also a highly genetic condition and parents likely are affected by symptoms such as disorganisation and impulsivity, which may impact on child sleep.

### Treating sleep problems in children with ADHD using behavioural strategies

We recently published our efficacy RCT evaluating the impact of a behavioural sleep programme in children with ADHD and moderate/severe sleep problems (n=244 children aged 5–12 years with ADHD).<sup>8</sup> Children were recruited from 21 paediatric practices across Victoria, Australia, and randomised to either a behavioural sleep intervention (n=122) or a usual care

comparison group (n=122). Intervention families attended 2× 50 min individual sessions (held 2 weeks apart) with a clinician trained in the intervention (psychologist or trainee paediatrician), comprising a standardised yet flexible intervention, tailored to the child's sleep problems and family preferences.

The intervention was associated with improved child outcomes at 3 and 6 months postrandomisation, including improved parent-reported child sleep, ADHD symptom severity, QoL, daily functioning and behaviour.<sup>8</sup> Intervention children also had improved teacher-reported classroom behaviour, as well as improved school attendance and working memory assessed via blinded assessment.<sup>8</sup> There were small improvements in sleep duration for a subsample that completed actigraphy.<sup>8</sup> Although the intervention was associated with improved caregiver work attendance and mental health 3 months later, these benefits were not observed at 6 months.<sup>8</sup>

Similarly, two additional RCTs have demonstrated the efficacy of improving sleep problems in children with ADHD. Keshavarzi *et al*<sup>9</sup> reported that a 12-week sleep training programme for children with ADHD aged 10 years (n=40) had beneficial effects for sleep and psychosocial functioning when compared with controls with ADHD who did not receive the intervention (n=20) and typically developing children (n=20). However, the professional group responsible for delivering the intervention was not reported. Corkum *et al*<sup>10</sup> also recently reported the beneficial effects of their distance sleep intervention delivered by paraprofessionals for childhood insomnia in a mixed sample of children with and without ADHD (n=61). Improvements were identified at 2 and 6 months for sleep assessed using parent report as well as actigraphy, and benefits were also reported for broader child psychosocial health.<sup>10</sup>

Although RCTs have shown that melatonin is associated with improved sleep onset latency for children with ADHD,<sup>5–7</sup> broader benefits for daily functioning including ADHD symptom severity have yet to be shown.

### Managing sleep problems in children with ADHD through existing clinical services

Although three trials have now shown that it is possible to improve sleep problems in children with ADHD using behavioural approaches, it remains to be seen whether it is possible to replicate such approaches and improve outcomes for children with ADHD in 'real life' clinical settings when delivered by their treating clinicians (eg, community paediatricians or psychologists) rather than by clinicians hired to deliver interventions in tightly controlled research trials. We are now testing the translation of this programme at the population level by training community paediatricians and psychologists to deliver this sleep intervention to children with ADHD and sleep problems. If this intervention can be translated successfully to the population level within existing workforces, then this would have the real potential to improve

outcomes for children with ADHD and their families. If cost-effective, it could be readily incorporated into the existing health system.

Paediatricians are the main care providers for ADHD in Australia.<sup>18</sup> The paediatrician's role includes the assessment and treatment (behavioural and medication-based) of ADHD and associated comorbidities (including sleep problems, if identified). Visits to a paediatrician are subsidised by Australia's Medicare scheme; however, families may experience out-of-pocket costs for these visits. Paediatricians (and general practitioners) may also refer children to a psychologist in the community to manage comorbid behaviours.<sup>19</sup> In Australia, this referral is facilitated by the Medicare Better Access to Mental Health Scheme, which provides subsidised access to up to 10 psychology sessions in a calendar year, provided the criteria for a DSM condition are met. We have deliberately designed our sleep intervention, so that it is feasible for paediatricians and psychologists to deliver within the Australian Medicare scheme and within their typical consultation durations.

### Aims and hypotheses

Building on our efficacy trial, we aim to determine whether the Sleeping Sound with ADHD intervention, delivered by paediatricians or psychologists in their usual work setting, can replicate the benefits of the efficacy trial. Therefore, in this translational, cluster-randomised trial, we aim to determine whether a brief sleep intervention delivered by paediatricians or psychologists:

1. Decreases the prevalence of children with moderate/severe sleep problems from moderate/severe to no/mild by parent report at 3 months postintervention (*primary outcome*).
2. Improves child and family functioning at 3 and 6 months postintervention (*secondary outcome*).
3. Is cost-effective (*secondary outcome*).

We hypothesise that, compared with the control children at 3 months (primary outcome timepoint) and 6 months postintervention, a brief behavioural sleep intervention will:

1. Decrease the prevalence of child sleep problems from moderate/severe to no/mild;
2. Improve functioning in other child (sleep severity, ADHD symptoms, QoL, behaviour, working memory, executive functioning, learning, academic achievement, school attendance) and primary caregiver (mental health, parenting, work attendance) outcome domains.
3. Be cost-effective.

## METHODS AND ANALYSIS

### Overall study design

This is a cluster RCT of a behavioural sleep intervention versus usual care conducted in Victoria and Queensland, Australia (see [figure 1](#)). The trial will be

reported according to Consolidated Standards of Reporting Trials guidelines and the extension report of non-pharmacological interventions.<sup>20</sup>

### Participants

Participants are parents of children aged 5–12 years at the time of recruitment with paediatrician-diagnosed and DSM-5 confirmed ADHD and a moderate to severe sleep problem as defined by the American Academy of Sleep Medicine diagnostic criteria:<sup>21</sup> chronic insomnia disorder, delayed sleep–wake phase disorder or sleep-related anxiety.

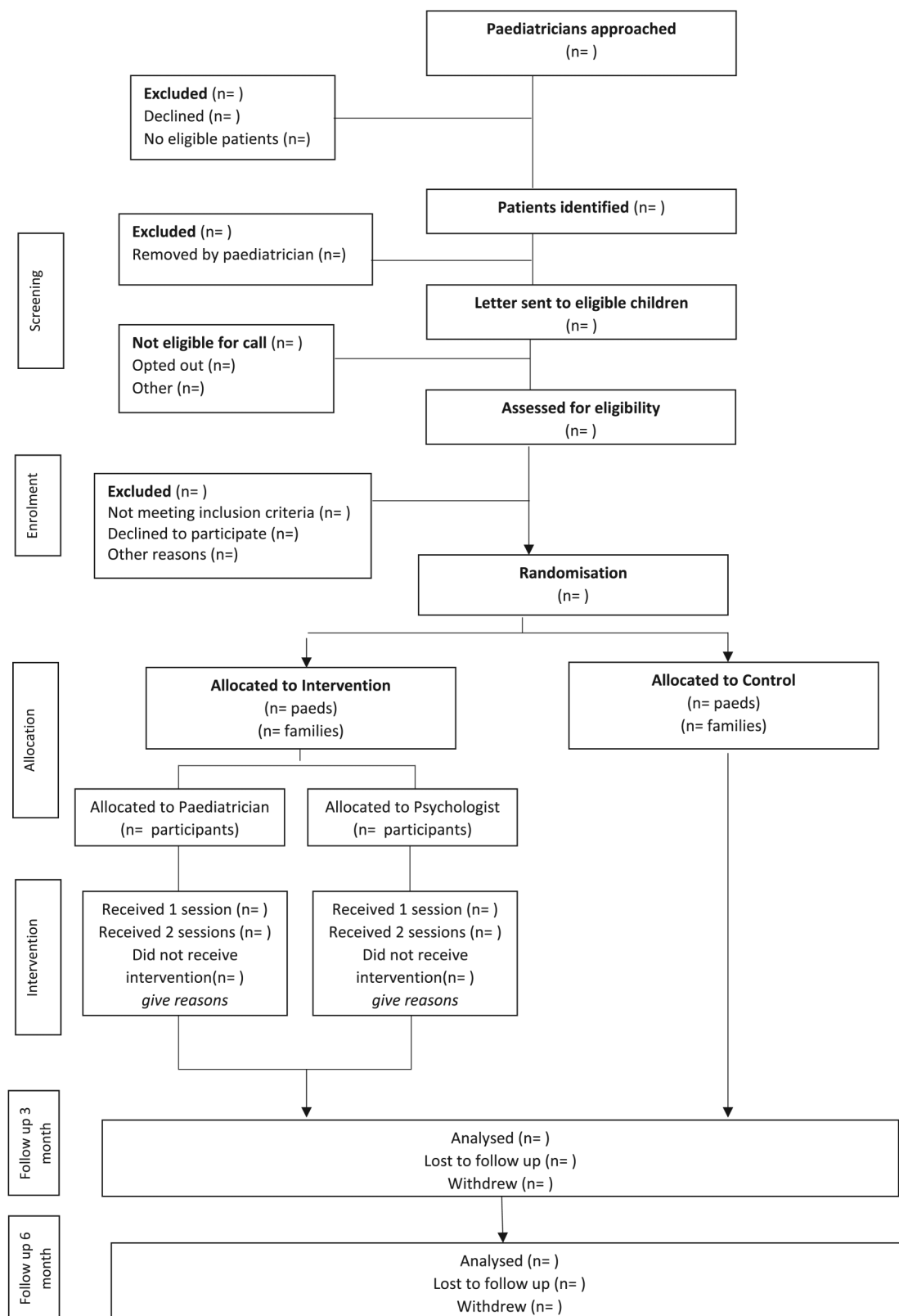
### Recruitment of health professionals

Given that paediatricians are the main healthcare provider for children with ADHD, recruitment and randomisation occurs at the level of the paediatrician. Paediatricians are recruited via the Australian Paediatric Research Network or through clinical contacts.<sup>18</sup> Interested paediatricians are invited to attend a briefing session in which the study requirements and timetable is presented and paediatricians have the opportunity to ask questions. Interested paediatricians are sent a follow-up email thanking them for their interest, asking them to sign an individual Memorandum of Understanding (MOU) if they consent to participation, and asking them to provide a list of psychologists to whom they refer patients. By signing the MOU, paediatricians agree to: (1) participate in training if randomised to the intervention arm; (2) deliver the sleep programme to intervention families *or* refer the child to a psychologist to deliver the sleep programme if allocated to the intervention; (3) not share the intervention materials with control group paediatricians if allocated to the intervention; and (4) allocate time for intervention session bookings.

Once paediatricians have been recruited, psychologists are invited to whom the paediatricians have indicated they refer patients. Psychologists are only recruited for the intervention arm of the project and have no contact with control participants. The research team then informs psychologists about the study via email or phone. The researchers then send psychologists who express interest in participating a follow-up email thanking them for their interest and asking them to sign an individual MOU, agreeing to: (1) participate in training; (2) deliver the sleep programme to intervention families referred by paediatricians; (3) use intervention materials only with children enrolled in the study; and (4) not share the intervention materials with any other families they see during the study period. This approach has worked well to minimise contamination in our previous RCTs.<sup>22–24</sup>

### Screening children for sleep problems and recruiting families

This translational trial uses, where possible, the same procedures that were used in our efficacy trial.<sup>8</sup>



**Figure 1** Participant flow.

Paediatricians preidentify their patients with ADHD, aged between 5 and 12 years, seen within the last 12 months, either through their medical software or through case notes. Paediatricians then send a letter to

the child's primary caregiver inviting them to take part. The letter advises families that the research team will ask them about their child's sleep and ADHD symptoms. An 'opt out' approach is used, whereby parents are asked to



contact the paediatrician if they *do not* wish to learn more about the study. If parents do not opt out within a 2-week period, the paediatrician provides the research team with the contact details of the families. Only 9% of families opted out in our efficacy trial.<sup>8</sup> Paediatricians also have the option to use an active consent process. In these cases, paediatricians send an 'opt in' version of the study invitation letter.

The research team then contacts interested (or not opted-out) families via telephone to ascertain if the child meets inclusion/exclusion criteria. Eligible families are sent a parent information statement and consent form (to participate in the RCT and for optional consents outlined below) and a baseline survey. Families have the option to complete hard copy surveys or online surveys (via REDCap—a secure, web-based application).<sup>25</sup> Surveys are completed by the child's primary caregiver. Parents have the option to consent to have their child's school teacher complete a survey about their child's behaviour. If consent is provided by the primary caregiver and school, a link to an online survey is sent to teachers. Parents also have the option to consent to being contacted for ethically approved future research, for the research team to keep their deidentified data to be shared with future ethically approved research, and to allow the research team to link with their child's *National Assessment Program—Literacy and Numeracy* (NAPLAN) results. NAPLAN is a national assessment of literacy and numeracy Australian children complete in Grades 3 and 5, and years 7 and 9.<sup>26</sup>

Once caregivers provide written consent, the research team contacts them to complete the Anxiety Disorders Interview Schedule for Children (ADIS-C) over the telephone to assess comorbid internalising and externalising diagnoses in children with ADHD.<sup>27</sup> To ensure we administer the telephone interviews consistently, we are recording 10% of the sample to test inter-rater reliability.

### Inclusion/exclusion criteria (all assessed via telephone)

#### Inclusion criteria

- Child aged between 5 and 12 years at the time of the recruitment call.
- Child's sleep is a moderate–severe problem for the caregiver ascertained using the following question: 'Has your child's sleep been a problem for you over the past 4 weeks?' If 'yes', they are asked to rate severity (mild, moderate or severe). This measure has been used in studies of children with and without ADHD and has good agreement with the Children's Sleep Habits Questionnaire.<sup>2 28 29</sup>
- Child sleep problem meets the International Classification of Sleep Disorders—Third Edition criteria<sup>21</sup> for an eligible sleep disorder (chronic insomnia disorder, delayed sleep–wake phase disorder) or sleep-related anxiety assessed using study-designed questions. To meet criteria for *chronic insomnia*

*disorder* in this study, children need to meet criterion A (one or more of the following: difficulty initiating sleep, difficulty maintaining sleep, waking earlier than desired, resistance going to bed, difficulty sleeping without a caregiver), B (evidence of impairments related to the sleep difficulty such as daytime sleepiness, impaired social, family, or academic performance, etc), C (sleep problem not be explained by inadequate sleep opportunity or circumstances) and D and E (symptoms and associated impairment must have been present three times a week or more for at least 3 months).<sup>21</sup> To meet criteria for *delayed sleep–wake phase disorder* in this study, criterion A (significant phase delay as shown by difficulty initiating sleep and awakening at a required time), B (delayed sleep–wake phase must have been present for at least 3 months) and C (improved sleep quality and duration when allowed to choose their own schedule) need to be satisfied.<sup>21</sup> Children were accepted into the study on the basis of experiencing sleep-related anxiety if the caregiver endorses significant difficulty falling asleep at night, in addition to anxious behaviour at bedtime (eg, fearful behaviours such as crying, asking for reassurance or lying in bed worrying).

- Child meets DSM 5 criteria for ADHD assessed via the 18-item ADHD Rating Scale IV—a validated scale measuring the core symptoms of ADHD.<sup>30</sup> Caregivers need to rate at least six of nine of the inattention and/or hyperactivity/impulsivity items as occurring 'often' or 'very often' in order to meet current symptom criteria. Caregivers are asked to rate symptoms of stimulant medication. Additional questions are then asked to ensure symptoms have been present for at least 6 months ('Did your child have these symptoms for at least 6 months before being diagnosed with ADHD?'), and contribute to cross-situational impairment ('Are these symptoms present at home, school and/or when out socially?'). Given the target age range, all participants will meet age of onset criteria. We do not assess whether the symptoms were better explained by another mental disorder.

Children are eligible to participate if they are taking melatonin or any other sleep-inducing medication, as long as the inclusion criteria are still met, despite medication use.

#### Exclusion criteria

- Suspected OSA identified using three OSA items from the Children's Sleep Habits Questionnaire.<sup>31</sup> Paediatrician investigators HHi and HHe telephone caregivers of children with suspected OSA to ask further about their symptoms and if OSA is suspected, these children are excluded and referred to appropriate clinical services as per usual clinical care (1% in our efficacy trial).<sup>8</sup>
- No longer meeting criteria for ADHD at the time of recruitment.

- C. Major illness or disability (eg, intellectual disability). Parents are asked if their child has ever been diagnosed with an intellectual disability or serious medical condition. If yes, parents are asked to provide further details (eg, IQ score, diagnosis). If clarification is needed, permission is sought to recontact the paediatrician to obtain relevant information from the child's medical record. Common comorbidities, including internalising/externalising disorder and autism spectrum disorder, are not excluded.
- D. Parents with insufficient English language proficiency to complete study documentation and/or participate in the intervention.
- E. Participated in our original efficacy trial.

### Sample size

Sample size planning is based on the primary outcome, the proportion of children for whom parents no longer report moderate/severe sleep problems at 3 months. In the efficacy study, at 3 months, 56% and 30% of parents in the intervention and control group, respectively, reported that their child's sleep was still a moderate/severe problem, that is, a difference of 26%. We conservatively assume a smaller difference of 20%, that is, 56% vs 36% to be observed in this trial. In order to have 80% power in detecting this difference at a two-sided type 1 error level of 5%, a group sample size of 107 individuals (total sample size  $n=214$ ) is required. As this is a cluster controlled trial (clustered at the level of the paediatrician to minimise contamination), we will inflate our sample size by a design effect of 1.2 where the design effect =  $1 + (n-1)r$  with  $n=3$  (the number of children seen by each paediatrician) and  $r=0.1$  (the conservatively assumed intracluster correlation coefficient, based on our efficacy RCT). Allowing for 20% drop out to 3 months and accounting for clustering, we need to recruit 160 children in each study arm (total sample size  $n=320$ ). All recruitment procedures described have been used in our previous trials to achieve similar recruitment numbers.

### Randomisation

Randomisation occurs at the level of the paediatrician. Paediatricians are randomly allocated to either receive the sleep training or not by an independent researcher, thereby avoiding potential contamination if all paediatricians were to be trained in the programme. Randomisation is based on a pregenerated group allocation sequence from the Murdoch Childrens Research Institute's Clinical Epidemiology and Biostatistics Unit and is stratified by location of the paediatrician (regional or metropolitan area). The randomisation is also stratified by the predicted number of enrolled patients (<10 and 10 or more). Paediatricians, families and researchers are blind to group allocation at the time of recruitment.

If the paediatrician is assigned to the intervention group and has more than four patients enrolled, families are randomised to receive the intervention from

either the paediatrician or psychologist. This second step of randomisation takes place after family recruitment is complete. The research team then sends the paediatrician a letter informing them of whether they will be receiving the sleep training or not. In addition, participating families are also sent a letter informing them of whether they will be receiving the sleep intervention or not.

Families of children attending a paediatrician randomised to the control group receive 'usual care' which involves seeing their paediatrician as per usual regarding their child's ADHD. Our research has shown that this does not routinely include addressing sleep issues.<sup>2</sup> In Australia, paediatricians typically see children with ADHD every 6 months to check their height, weight, blood pressure and reissue a script for medication (valid for 6 months), where necessary.

### Intervention training

The intervention is designed to be feasible to deliver in paediatricians' and psychologists' practice. Intervention group paediatricians and psychologists are trained to deliver the same programme described for our efficacy trial<sup>8</sup> in one 3.5 hour session. Training occurs conjointly for paediatricians and psychologists, where possible, with investigators HHi, ES and HHe to maximise fidelity. Training addresses normal sleep patterns in school-aged children and highlights the importance of sleep hygiene practices such as consistent bedtime routines and keeping bedrooms media-free. The intervention includes verbal and written information of standard sleep intervention strategies as recommended by the American Sleep Association<sup>32</sup> and addresses common sleep problems experienced at the beginning of, and during, the night. A summary of sleep problems addressed and management strategies are provided in table 1. Training focuses on how these strategies can be individualised for families and includes evidence-based instructional strategies such as role play, feedback, modelling practice and use of checklists. Training is supported by a manual and written parent education materials.

Prior to training, paediatricians and psychologists complete a 'pre-quiz' to establish their baseline knowledge, skills and attitudes in relation to child sleep problems and their management. A 'post-quiz' following training is used to assess changes in knowledge, confidence and perceived competence in the management of child sleep problems.

### Intervention delivery

Paediatricians and psychologists are sent an intervention package for each child allocated to them containing the child's contact details, a clinical checklist to complete after each appointment and parent education materials. Families contact the clinician they have been allocated to make an appointment (up to 30 min for paediatricians, up to 50 min for a psychologist) to discuss their child's sleep, with a follow-up session (up to 30 min for

**Table 1** Key behavioural sleep management strategies

Sleep disorder	Definition	Examples of behavioural strategies
Sleep-onset association disorder	Child associates falling asleep with a person or object (eg, television) and is unable to fall asleep without its presence	<ul style="list-style-type: none"> <li>▶ Adult fading (ie, graduated extinction) using 'camping out'—gradual withdrawal of parental presence from the child's bedroom over 7–10 days</li> <li>▶ 'Checking method'—parent checks on the child at regular time intervals (2, 5 or 10 min, with intervals increasing over time)</li> </ul>
Delayed sleep phase	Shift in the child's sleep–wake cycle, in which the child cannot fall asleep until late and then wakes late in the morning	<ul style="list-style-type: none"> <li>▶ Bedtime fading—child's bedtime is temporarily set later to when they are usually falling asleep and gradually brought forward. The child is then woken at a preset time in the morning</li> <li>▶ Early morning light exposure</li> </ul>
Limit setting sleep disorder	Child refusal to go to bed and general non-compliant behaviour at bedtime. Parent struggles to set appropriate and consistent bedtime limits	<ul style="list-style-type: none"> <li>▶ Parent management strategies—ignoring child protests, rewarding compliance with bedtime routines. A 'bedtime pass', whereby the child can only leave the bedroom one time before sleep, can be used to promote compliant behaviour</li> <li>▶ Consideration of bedtime fading or the checking method</li> </ul>
Primary insomnia	Child has substantial difficulty initiating and/or maintaining sleep even if they go to bed at a later time	<ul style="list-style-type: none"> <li>▶ Visual imagery and relaxation</li> <li>▶ Basic cognitive restructuring</li> <li>▶ Restricting time in bed (eg, temporarily setting the bedtime later as per delayed sleep phase or getting out of bed and doing a relaxing activity if the child cannot sleep)</li> </ul>
Night-time anxiety	Specific night-time fears including fear of the dark and/or child worrying about other things while in bed	<ul style="list-style-type: none"> <li>▶ Visual imagery and relaxation training</li> <li>▶ Discussing fears during the day rather than just before bedtime</li> <li>▶ Rewarding brave behaviour</li> <li>▶ Other—use of a security object, avoiding scary television shows, use of a book to record worries</li> </ul>

paediatricians, up to 50 min for a psychologist) ~2 weeks later and if needed, a final 15–20 min phone call a further 2 weeks after that. The differences in consultation duration reflect real-life clinical practice. We record the actual duration of all consultations, which will enable us to examine the relationship between intervention dose and outcomes. Children are present at all face-to-face appointments. The first session focuses on an assessment of the child's sleep problem, providing information about normal sleep and sleep cycles, giving the family a sleep diary to complete, advice about sleep hygiene and a plan specifically tailored to the child's particular sleep disorder. The second session and follow-up phone call include a review of the sleep diary, re-enforcement/trouble shooting of existing strategies and the introduction of new strategies, where appropriate.

The research team contacts families within 1 month postrandomisation to ensure an appointment has been made with either their paediatrician or a psychologist and to encourage them to make the appointment if they have not yet performed so.

### Follow-up

We mail/email follow-up surveys to families and teachers at 3 and 6 months postintervention (measured from first intended intervention session for intervention families

with controls followed up at the median date of intervention follow-up) to determine short and longer term changes in primary and secondary outcomes. Teachers are not informed of the child's intervention group status. For intervention families only, the 3-month survey also asks about the behavioural intervention components received, usefulness of the intervention, ease of use of strategies, costs experienced by families in accessing the programme and any negative events/experienced. At 6 months, a trained research assistant completes a blinded direct assessment of child executive functioning, learning, working memory, academic functioning, sleep and QoL via home visits (of 45 min duration).

### Measures

All measures administered at baseline, 3 and 6 months are summarised in table 2. The majority of measures were used in our efficacy trial. We now additionally measure academic functioning, given that our efficacy trial showed a trend towards improvement in working memory, which is a key determinant of academic functioning. For similar reasons, we also include a more detailed assessment of cognitive and executive functioning. The baseline questionnaire includes family (eg, family composition, parental education and age, language spoken at home, annual household income) and

**Table 2** Summary of measures

Construct	Measures	Source	T1	T2	T3
Child outcomes					
Sleep	<i>Sleep problem prevalence (primary outcome)</i> —Primary caregiver report of child sleep problem (none/mild vs moderate/severe) <sup>2</sup>	P	●	●	●
	<i>Children's Sleep Habits Questionnaire (CSHQ)</i> —33-item validated measure of disorders of initiating and maintaining sleep. <sup>31</sup> Child Sleep Self-Report collected at 6 months only <sup>15</sup>	P	●	●	●
	<i>Teacher Daytime Sleepiness Questionnaire</i> —10-item validated scale of daytime sleepiness at school <sup>39</sup>	T	●	●	●
	<i>Sleep Hygiene scale</i> —7-item measure assessing sleep hygiene <sup>24</sup>	P	●		●
Comorbidity	Anxiety Disorders Interview Schedule for DSM-IV ( <i>ADIS-C</i> )—diagnostic interview assessing mental health disorders according to DSM-IV criteria. This is completed with parents over the telephone and has been validated for this purpose <sup>27</sup>	P	●		
ADHD	<i>ADHD Rating Scale IV</i> —18-item validated scale measuring the core symptoms of ADHD <sup>30</sup>	P, T	●	●	●
Behaviour	<i>Strengths and Difficulties Questionnaire</i> —25 items assessing the following subscales: hyperactivity/inattention, conduct problems, emotional symptoms, peer relationship problems, and prosocial behaviour <sup>40</sup>	P, T	●	●	●
Irritability	<i>Affective reactivity index (ARI)</i> —6-item validated measure assessing child irritability <sup>41</sup>	P	●	●	●
School attendance	<i>School attendance</i> over the preceding 3 months <sup>2</sup>	P	●	●	●
Quality of life	<i>Child Health Utility-9D (CHU-9D)</i> —9-item measure of child quality of life used to calculate quality-adjusted life years. Child report is collected at the 6-month follow-up only <sup>42</sup>	P, C	●		●
	<i>Pediatrics Quality of Life Inventory 4.0</i> —23-item measure of child total, physical and psychosocial QoL <sup>43</sup>	P	●		●
Memory	<i>Sleep Suite App</i> —An animated iPad application that includes a continuous performance test to measure sustained attention, and 2 tests of memory consolidation and learning <sup>44</sup>	C			●
Working Memory	<i>Working Memory Test Battery for Children</i> —The Backward digit span recall subtest is administered as a measure of child working memory <sup>45</sup>	C			●
Academic functioning	<i>Wide Range Achievement Test 4 (WRAT)</i> —word reading and math computation subtests <sup>46</sup>	C			●
	<i>NAPLAN</i> —standardised academic tests of reading, writing, language conventions and numeracy. Results can be compared with National or State population results and against the national minimum standard <sup>26</sup>	L			●
Autism	<i>Social Responsiveness Scale (SRS) Brief</i> —16-item measure of autism spectrum disorder symptoms <sup>47</sup>	P			●
Parent outcomes					
Mental health	<i>Kessler 6 (K6)</i> —6-item validated measure of adult psychological stress <sup>48</sup>	P	●	●	●
Work attendance	<i>Work attendance</i> over the previous 3 months <sup>2</sup>	P	●	●	●
Parenting	<i>Parenting</i> —validated scales developed for the Longitudinal Study of Australian Children assessing parenting consistency (6 items) and parental warmth (5 items) <sup>49</sup>	P	●		●
Family costs	<i>Service use and costs</i> —families report service use over previous 3 months as well as time, travel and out-of-pocket costs associated	P		●	●

C, child-report; L, data linkage; P, parent-report; T, teacher-report; T1, baseline; T2, 3 months postintervention; T3, 6 months postintervention.

child demographic and medical (eg, medication, diagnosed comorbidities, etc) characteristics.

The research team prospectively records resources used to administer all programme components to facilitate economic analyses. Families also report time, travel and out-of-pocket costs associated with visits as well as those costs associated with child ADHD medication/prescriptions.

### Data analysis

Primary (aim 1) and secondary (aim 2) analyses will be based on the 'intention to treat' population at the level of the individual child and will adjust for clustering at the level of the paediatrician using a generalised linear mixed effect model (logit link function) for the binary primary outcome (proportion of children with moderate/severe sleep problems). For primary and secondary



analyses, the stratification variable 'location of study site (practice)' as well as an indicator variable for the type of healthcare professional who delivered the intervention (paediatrician or psychologist) will be considered as adjustments variables. For all analyses, model-based effect estimates (risk differences and risk ratios) will be reported along with 95% CIs. Because of the range of outcomes and timepoints, the results will be considered in their totality and interpreted with suitable caution regarding formal conclusions of 'statistical significance'. In order to reduce potential confounding bias due to differential patient-drop out over time, multiple imputation methodology will be considered to replace missing outcome data in the primary efficacy analysis. All secondary analyses (aim 2) will be adjusted for baseline scores and stratification variables wherever possible, in order to increase precision of comparisons.<sup>33</sup> For primary and secondary analyses, we will present results of analyses unadjusted and adjusted for further potential outcome predictor variables (child age, medication use, comorbidities, and family sociodemographic characteristics (Socioeconomic Index for Advantage linked to the family's postcode),<sup>34</sup> parent high school completion and parent age). Syntax will be written before database lock, which will cover the primary and secondary analyses. Participant-level data are available on request.

### Cost-effectiveness analyses

The economic analysis (aim 3) will first present cost-consequences analysis, then proceed to cost-effectiveness analysis against the primary outcome (*prevalence of sleep problems*) measure as the primary economic analysis. Secondary economic analyses will include cost-utility analysis against CHU9D-based QALYs.<sup>35 36</sup> These analyses will be conducted from the healthcare and broader societal perspectives (as cost-effectiveness can vary significantly by perspective)<sup>37</sup> and will include extensive sensitivity analyses to test the impact of uncertainty in data and sensitivity to evaluation approach.

### DISSEMINATION

ADHD affects ~5% of school-aged children<sup>38</sup> and is frequently associated with sleep problems, which worsen child and family outcomes.<sup>2</sup> Findings will determine whether the benefits of an efficacy trial can be realised more broadly at the population level and whether it is cost-effective to do so. If cost-effective, we expect the following outcomes: (1) the best evidence yet that addressing sleep problems improves outcomes for children with ADHD; and (2) a ready-to-use intervention that is tailored to the Australian health system, and can be replicated internationally. Findings will inform the development of clinical guidelines for managing sleep problems in this population. We will seek to publish in leading international paediatric journals, present at major conferences and disseminate findings through established clinician networks.

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**Patient consent** Obtained.

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