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Behavioural sleep problems in children with attention-deficit/hyperactivity disorder (ADHD): protocol for a prospective cohort study

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ABSTRACT

Introduction: Children with attention-deficit/hyperactivity disorder (ADHD) commonly experience behavioural sleep problems, yet these difficulties are not routinely assessed and managed in this group. Presenting with similar symptoms to ADHD itself, sleep problems are complex in children with ADHD and their aetiology is likely to be multifactorial. Common internalising and externalising comorbidities have been associated with sleep problems in children with ADHD; however, this relationship is yet to be fully elucidated. Furthermore, limited longitudinal data exist on sleep problems in children with ADHD, thus their persistence and impact remain unknown. In a diverse sample of children with ADHD, this study aims to: (1) quantify the relationship between sleep problems and internalising and externalising comorbidities; (2) examine sleep problem trajectories and risk factors; and (3) examine the longitudinal associations between sleep problems and child and family functioning over a 12-month period.

Methods and analysis: A prospective cohort study of 400 children with ADHD (150 with no/mild sleep problems, 250 with moderate/severe sleep problems) recruited from paediatric practices across Victoria, Australia. The children’s parents and teacher provide data at baseline and 6-month and 12-month post enrolment.

Key measures: Parent report of child’s sleep problem severity (no, mild, moderate, severe); specific sleep domain scores assessed using the Child Sleep Habits Questionnaire; internalising and externalising comorbidities assessed by the Anxiety Disorders Interview Schedule for Children IV/Parent version.

Analyses: Multiple variable logistic and linear regression models examining the associations between key measures, adjusted for confounders identified a priori.

Ethics and dissemination: Ethics approval has been granted. Findings will contribute to our understanding of behavioural sleep problems in children with ADHD. Clinically, they could improve the assessment and management of sleep problems in this group. We will seek to publish in leading paediatric journals, present at conferences and inform Australian paediatricians through the Australian Paediatric Research Network.

Strengths and limitations of this study

▪ The sample is recruited from a geographically diverse population of children with attention-deficit/hyperactivity disorder and represents the case mix seen in clinical practice, thus improving generalisability of the findings.
▪ The longitudinal nature of the study will allow sleep trajectories to be identified in children with ADHD.
▪ Limitations of this study include lack of direct assessments of child’s sleep and exclusion of non-English speaking families.

BACKGROUND

Attention-deficit/hyperactivity disorder (ADHD) is a heterogeneous disorder that leads to impairment across multiple domains including social and academic functioning.1 ADHD affects up to 5% of children worldwide2 and is commonly accompanied by sleep problems.3 Between 55% and 74% of parents report sleep problems in their child with ADHD4–8 and these are typically behavioural in nature (ie, extrinsic cause) and can include difficulty falling asleep, night-waking and bedtime resistance.9–9 Primary sleep disorders (ie, intrinsic cause) such as restless leg syndrome and sleep disordered breathing are also elevated in children with ADHD compared with typically developing children.9–12 Yet, despite a longstanding recognition of sleep problems prevalence,13,14 and evidence that they are associated with poorer child and family well-being,6 sleep problems do not appear to be routinely assessed or managed by clinicians caring for children with ADHD.6 Furthermore, evidence-based treatments for sleep disturbances in children with ADHD remain limited.15

Identifying sleep problems in children with ADHD is challenging. Sleep deprivation closely mimics symptoms attributed to ADHD...
(eg, inattention and hyperactivity)10 16 and objective and subjective measures of sleep often have disparate findings.4 16 17 Objective measures (eg, polysomnography and actigraphy)—the ‘gold standard’—produce reliable findings for parameters such as sleep duration18 but feasibility and costs are often barriers to their use in large studies.19 In addition, they do not capture behavioural sleep problems such as bedtime resistance and limit setting disorders—which are more common in children with ADHD. On the other hand, subjective measures (eg, parent report) are a valid screening tool for behavioural sleep disorders18 yet may overestimate sleep duration, underestimate night-waking,18 19 and may also be influenced by parental mental health.20 Parent report is, however, a valid measure for large studies where behavioural sleep problems are the key focus. Furthermore, parent report reflects parental concern, which is a key driver for seeking health professional advice around sleep.21

The mechanisms associated with sleep problems in children with ADHD are complex and appear multifactorial.10 Sleep problems and ADHD are regulated by the central nervous system, thus there is a strong neurobiological overlap3 16 22 and their relationship is likely bidirectional.10 25 Empirically, this is supported by the recognition that ADHD symptom severity worsens in the presence of sleep problems.6 24 25 Similarly, stimulant medications (eg, methylphenidate), which also work on the central nervous system, have been associated with sleep problems, in particular delayed sleep-onset latency.13 20–22 Yet, in some circumstances, stimulants may improve sleep26 and many stimulant naïve children also experience sleep problems,5 therefore other factors have received increasing investigation.

In typically developing children, sleep problems have been associated with internalising (eg, mood disorders and anxiety disorders) and externalising (oppositional defiant disorder, conduct disorder) comorbidities.27 Research suggests that this is also the case for children with ADHD.24 25 28–30 However, this relationship is yet to be fully elucidated. Previous studies are often limited to single clinical samples5 24 25 31 32 and exclude children with common comorbidities or those taking ADHD medication.28 29 33 Furthermore, researchers are yet to consider children with co-occurring internalising and externalising comorbidities. Elucidating the relationship between sleep problems and internalising and externalising comorbidities in children with ADHD is likely to inform clinical practice around the assessment and management of sleep problems in this group.

Clinical management of sleep problems is also restricted by the lack of longitudinal data, which would inform the prognosis and thus the relative importance of managing sleep problems in children with ADHD. Although behavioural sleep problems are common in typically developing children, they are often transient and likely to resolve naturally.34 However, in children with major depression, sleep problems have been shown to persist even after successful treatment.35 The nature and persistence of sleep problems in children with ADHD is unclear. In a large population-based longitudinal study, Scott et al23 showed that children with ADHD slept less than typically developing children from infancy to age 11 years, and this was statistically significant at ages 5.9, 6.9 and 9.7 years. This study has made a substantial contribution to the literature, yet its focus is limited to sleep duration and does not consider other common sleep problems. In a longitudinal study of a clinical sample of children referred with ADHD, anxiety or both (n=76, ages 7–13 years), sleep problems were more likely to persist in this group over an 18-month period compared with a control group of children without a history of accessing mental health services recruited from nearby schools (n=31).36 The generalisability of these findings is limited by the small sample size and mixed sample of children with anxiety alone and ADHD/anxiety. Longitudinal studies that can be generalised to the wider population of children with ADHD are required to understand trajectories and prognosis of sleep problems in ADHD.

Furthermore, longitudinal data could assess the burden of sleep problems in ADHD. Cross-sectionally, children with ADHD and parent-reported sleep problems have higher ADHD symptom severity24 25 37; poorer quality of life, daily functioning and school attendance and their caregivers are also more likely to miss work and have poorer mental health6 compared with children with ADHD without sleep problems. However, the cross-sectional nature of these studies makes it impossible to delineate the sequence of sleep problems and other difficulties with functioning. Identifying the long-term impact sleep problems have on the functioning of children with ADHD is important to rationalise time and financial investment in the identification and management of sleep problems by health professionals and families.

**STUDY AIMS**

Through establishing a diverse cohort of children with ADHD recruited from multiple clinical practices, our study aims to address some of the previous limitations identified. The primary aim of the study is to examine associations between sleep problems and internalising and externalising comorbidities, considering parent-reported moderate/severe behavioural sleep problems (aim 1a) and sleep problem domains (eg, bedtime resistance and sleep anxiety) (aim 1b). Through longitudinal study of the cohort over a 1-year period, we will examine the trajectories of behavioural sleep problems and predictive risk factors (aim 2) and examine longitudinal relationships between behavioural sleep problems and child and family well-being, specifically in the domains of child quality of life, school attendance, parent mental health, family functioning and work attendance (aim 3).
METHODS AND ANALYSIS

Our prospective cohort study of children with ADHD (see figure 1) runs from April 2011 to July 2014, encompassing participant recruitment, baseline and follow-up data collection and data analysis.

Study procedure
Paediatric practice recruitment
Personalised emails are sent to paediatricians working in public and private practice inviting them to assist with participant recruitment. In Australia, many general paediatricians work in public and private practice and the most common condition they manage is ADHD.48 We identified paediatricians through the Australian Paediatric Research Network48 and by our knowledge of paediatricians who treat children with ADHD in the state of Victoria. Paediatricians preidentify children with ADHD who have been seen within the past 12 months. An ‘opt out’ letter, signed by the paediatrician, is sent to the child’s primary caregiver, asking them to return the letter to the paediatrician if they do not wish to be contacted about the study. If caregivers do not ‘opt out’ within a 2-week period, paediatricians pass the family’s contact details onto the research team. This method has previously resulted in good uptake rates.39

Family recruitment
The research team contacts families through telephone to assess eligibility. Interested, eligible families are mailed a participant information statement, consent form and parent baseline questionnaire. Families are enrolled on receipt of written informed consent and the baseline questionnaire. To determine the extent of participation bias, data collected will be compared with non-participants including child’s gender, age and socioeconomic status of the family’s immediate neighbourhood.

Participants
Inclusion criteria
Eligible children are aged 5–13 years who have a previous ADHD diagnosis, are currently experiencing ADHD symptoms and whose caregiver can report on their sleep, as described below.

Attention-deficit/hyperactivity disorder
Children identified by their treating paediatrician will have a confirmed diagnosis of ADHD, which in accordance with national recommendations requires confirmation of cross-situational impairment at kindergarten or school as well as home.40 ADHD symptoms must also be current and are, therefore, assessed during the recruitment call, using the validated 18-item ADHD Rating Scale (DSM-IV)41 and study-designed questions assessing ADHD diagnosis as per DSM-IV.42 The study designed questions include: (1) symptom duration (Did your child have these symptoms for 6 months or longer before he/she was diagnosed with ADHD?); (2) age of onset (Did your child have these symptoms before he/she turned seven?) and (3) cross-situational impairment (Are these symptoms present at home, school or when out socially (eg, in the park, visiting friends?).

Sleep problem is determined by parent report of their child’s sleep; parents are asked “Has your child’s sleep been a problem for you over the past 4 weeks?” If the parent responds ‘yes’, they are asked to rate the severity of the problem (mild/moderate/severe).6 34 43 In typically developing children, of those whose parents report a moderate/severe sleep problem, 90% fall in the clinical range on the validated Children’s Sleep Habits Questionnaire (CSHQ).44 Furthermore, parent report of moderate/severe sleep problems is associated with poorer child learning by blinded teacher report66 and poorer child and family well-being.9 During the recruitment call if parents report that their child has a moderate/severe sleep problem they are asked several questions to establish whether the sleep problem meets the International Classification of Sleep Disorders criteria for at least one behavioural sleep disorder45 or corresponds with night-time anxiety. Children with a moderate/severe sleep problem that meets at least one of these criteria are eligible for the Sleeping Sound with ADHD study, a randomised controlled trial (RCT) investigating whether treating sleep problems in children with ADHD can reduce ADHD symptoms.46 Children with no/mild sleep problems are eligible for the Attention to Sleep study, an observational cohort study.

Exclusion criteria
Children are excluded at the recruitment call if their parents report any of the following criteria:

1. Receiving specialised help for their sleep from a psychologist or a specialised sleep clinic—an important consideration for determining the efficacy of the intervention in the RCT;
2. Serious medical condition (eg, severe cerebral palsy) or an intellectual disability (IQ<70);
3. Suspected obstructive sleep apnoea (OSA). OSA is assessed using the three sleep disordered breathing items from the CSHQ.47 Parents who report that their child ‘sometimes or usually snores loudly’, ‘stops breathing’ and/or ‘snorts/gasps’ during their sleep are contacted by a paediatrician (HH) for further assessment. If OSA is suspected, children are referred to appropriate services;
4. Family does not speak sufficient English to complete recruitment call or questionnaires.

Data collection
Immediately post enrolment, families are sent a 7-day sleep and medication log to complete during a typical school week and scheduled for a phone interview to assess child internalising and externalising comorbidities (described below). If the primary caregiver consents, the child’s teacher is also sent a brief survey to assess classroom behaviour. Teachers are not informed of the
Follow-up surveys are sent to the child’s primary caregiver and teacher at 6-month and 12-month post enrolment (see table 1). Our follow-up period was limited to 12 months in order to harmonise with RCT data collection.  

### Measures

**Internalising and externalising comorbidities** are assessed by the Anxiety Disorders Interview Schedule for Children/Parent version IV (ADIS-C). The ADIS-C is a semi-structured interview that allows, ‘for research purposes, reliable and valid diagnosis’ according to DSM-IV criteria. The ADIS-C is validated for administration over the telephone with parents—an important practical consideration—and yields excellent to acceptable 7–14-day test-retest reliability estimates for parents (k=0.65–1.00). We assess 11 ADIS-C domains including: separation anxiety, social phobia, specific phobia, panic disorder, generalised anxiety disorder, obsessive compulsive disorder, post-traumatic stress disorder, dysthymia, major depressive disorder, conduct disorder and oppositional defiant disorder. Screening positive for two or more anxiety problems on a similar measure has been shown to (1) have high sensitivity (0.75) and a low false-positive rate (0.33) and (2) be associated with a fivefold increase in seeking treatment for an anxiety disorder. Thus, children screening positive for two or more anxiety problems on the ADIS-C or one mood disorder are classified as having an internalising comorbidity. Children screening positive to oppositional defiant or conduct disorder are classified as having an externalising comorbidity.

All researchers administering the ADIS-C hold at least a four-year degree in psychology and had received training and supervision from experienced clinical psychologist (ES). As interviewers are unblind to child’s sleep problem severity, approximately 10% of interviews are recorded to ensure reliability.

*These children had a moderate/severe sleep problem by parent report but it did not meet the International Classification of Sleep Disorders criteria for at least one behavioural sleep disorder or correspond with night-time anxiety.*
(with verbal consent) to allow blinded cross-coding in order to calculate inter-rater reliability coefficients.

Table 1 summarises all other key outcome and exposure measures collected during the 12-month period. We also collect data on child demographics, medication use, as well as parent age and education level, and neighbourhood socioeconomic disadvantage score—measured by the census-based Socio-Economic Indexes for Areas Disadvantage Index for the child’s postcode of residence (national mean 1000, SD 100; higher scores reflect greater advantage).52

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Measures collected across time points</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Measures</strong></td>
<td><strong>Baseline</strong></td>
</tr>
<tr>
<td>Child sleep/medication</td>
<td></td>
</tr>
<tr>
<td>Children’s Sleep Habits Questionnaire47</td>
<td>•</td>
</tr>
<tr>
<td>7-day sleep/medication log</td>
<td>•</td>
</tr>
<tr>
<td>Child behaviour and quality of life</td>
<td></td>
</tr>
<tr>
<td>Daily parent rating of evening/morning behaviour44</td>
<td>•</td>
</tr>
<tr>
<td>ADHD IV Rating Scale (parent and teacher versions)41</td>
<td>• ▲</td>
</tr>
<tr>
<td>Strengths and Difficulties Questionnaire55</td>
<td>• ▲</td>
</tr>
<tr>
<td>Pediatric Quality of Life Inventory—4.056</td>
<td>•</td>
</tr>
<tr>
<td>School attendance6</td>
<td>•</td>
</tr>
<tr>
<td>Parent/family outcomes</td>
<td></td>
</tr>
<tr>
<td>Work attendance6</td>
<td>•</td>
</tr>
<tr>
<td>Depression Anxiety Stress Scale57</td>
<td>•</td>
</tr>
<tr>
<td>Child Health Questionnaire: Family Impact Scale68</td>
<td>•</td>
</tr>
</tbody>
</table>

• Parent; ▲ Teacher; ADHD, attention-deficit/hyperactivity disorder.*

Sample size
The primary aim (1a) is to compare the proportion of co-occurring internalising and externalising comorbidities between children with moderate/severe and no/mild sleep problems. The established Sleeping Sound with ADHD RCT will provide data for 250 children with
ADHD and moderate/severe sleep problems. Initial baseline data from the trial (n=64) demonstrated high ADIS-C completion rates (90%) and high proportions of co-occurring internalising and externalising comorbidities (42%). To detect a clinically meaningful difference of 15% in the proportion of co-occurring internalising and externalising comorbidities between children with moderate/severe sleep problems (42%) and no/mild (27%) sleep problems, a further 150 children with no/mild sleep problems are required in the observational cohort study. Thus, our total study sample size is 400 children, which provides 80% power at a 5% (two-sided) level of significance and allows for 10% of parents not completing the ADIS-C.

**Data analysis**

Aims 1a and 1b involve analysis of baseline data only, and thus will include the full sample of 400 children. For aims 2 and 3, which include longitudinal data analyses, we will exclude children in the intervention arm of the trial (n=125) because of the expected effects of intervention on children’s sleep. Thus, the longitudinal sample includes ~125 children with moderate/severe sleep problems from the control arm and 150 children with no/mild sleep problems.

To address aim 1a, χ² tests will compare proportions of co-occurring internalising and externalising comorbidities between children with moderate/severe sleep problems and those with no/mild sleep problems. Children will then be classified into four groups: (1) ADHD alone (reference group), (2) internalising comorbidity alone, (3) externalising comorbidity alone and (4) both comorbidities (ie, internalising and externalising). Logistic regression will compare the odds of a moderate/severe sleep problem for children with each comorbidity type with the reference group. The model will adjust for potential confounders identified a priori including child age, gender, family sociodemographic factors, use of stimulant medication and ADHD symptom severity. Sensitivity analyses will be conducted excluding children who (1) are taking sleep medications, which could treat the sleep problem but not address comorbidities; (2) no longer meet criteria for an internalising comorbidity once sleep symptoms are excluded from the diagnostic criteria—to ensure that internalising comorbidities are not endorsed because of the child’s sleep problems when they would not be otherwise and (3) including parent mental health (ie, total Depression Anxiety Stress Scale score) in the adjusted model to examine the influence of parent mental health which could potentially be an outcome or a risk factor for comorbidities and sleep problem severity.

To address aim 1b, associations between comorbidities and sleep problem domains identified by the CSHQ, linear regression will estimate the mean change in domain scores for children with each type of comorbidity compared with ADHD alone (reference group).

Unadjusted and adjusted models controlling for confounders (outlined above for 1a) will be conducted.

To address aim 2, trajectories of sleep problems over the 12-month period, parent report of child’s sleep problems across the three time points (baseline, 6-month and 12-month follow-up) will be characterised including timing and persistence of sleep problems. At each time point, sleep problems will be dichotomised as no/mild versus moderate/severe (for the purpose of this analysis, no/mild will be referred to as ‘no sleep problem’ and moderate/severe as ‘a sleep problem’). It is anticipated that the categories derived will include the following sleep problem groups: (1) never; (2) remitted (no longer a problem); (3) incident (developed problem over the 12 months); and (4) persistent (problem at all three time points).

Using these categories as a nominal longitudinal outcome, multinomial logistic regression will examine baseline risk factors (ie, internalising and externalising comorbidities, ADHD symptom severity, ADHD and sleep medication use, family sociodemographic factors, age, gender, autism spectrum disorders and parent mental health).

To address aim 3, child and family well-being at 12 months will be examined in relation to the patterns of sleep problems experienced over the preceding 12 months. Child and family outcomes will include parent-reported child quality of life; parent-reported and teacher-reported child emotional, behavioural and social problems; parent report of school attendance; parent mental health, family functioning and work attendance. Linear and logistic regression models will be specified according to the outcome and each sleep problem trajectory will be included as a nominal predictor variable. Depending on the outcome examined, we will adjust for child factors including age, gender, ADHD and sleep medication use and family factors including sociodemographic factors. For outcomes other than parent mental health, this will additionally be considered as a confounding factor in sensitivity analyses. Unadjusted and adjusted regression models will be interpreted to consider how sleep problem trajectories may affect child and parent outcomes following a causal logic framework.

**DISSEMINATION**

Our study will elucidate the relationship between sleep problems and internalising and externalising comorbidities, which is likely to lead to better clinical identification of sleep problems and associated comorbidities. It will provide the first longitudinal data on sleep problems that are generalisable to the wider population of children with ADHD. These data will inform knowledge about the trajectories, risk factors and the impact of sleep problems in children with ADHD, which will be highly clinically relevant. If sleep problems are persistent for children with ADHD and associated with poorer outcomes longitudinally, then sleep interventions could provide a mechanism to improve outcomes for these children who already face considerable burden in our
society. Limitations of this study include lack of direct assessments of child’s sleep and exclusion of non-English speaking families. The study also has a number of important strengths, including the large and geographically diverse sample and strong, standardised measures. We plan to present our findings at conferences and publish in international peer-reviewed journals. We will also inform Australian paediatricians—the main care providers for children with ADHD—through the Australian Paediatric Research Network.

Contributors KL, HH, ES and FM conceived the study. KL coordinated the study and drafted the current manuscript, supervised by HH, ES and FM. AG participated in the coordination of the study and drafted the current manuscript, supervised by KL. All authors contributed, read and approved the final manuscript.

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Competing interests None.

Ethics approval The study is approved by The Royal Children’s Hospital Melbourne Human Research Ethics Committee (30033, 28017) and Victorian Department of Education and Early Childhood Development (000573, 001307).

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