



Knight, F., & Dimitriou, D. (2016). Methodologies for paediatric sleep research in typical and atypical populations. In J. Prior, & J. Van Herwegen (Eds.), *Practical Research with Children* (pp. 246-263). Routledge.

Peer reviewed version

[Link to publication record on the Bristol Research Portal](#)
PDF-document

This is the author accepted manuscript (AAM). The final published version (version of record) is available via Routledge . Please refer to any applicable terms of use of the publisher.

University of Bristol – Bristol Research Portal

General rights

This document is made available in accordance with publisher policies. Please cite only the published version using the reference above. Full terms of use are available:
<http://www.bristol.ac.uk/red/research-policy/pure/user-guides/brp-terms/>

13. Methodologies for paediatric sleep research in typical and atypical populations

Frances Le Cornu Knight and Dagmara Dimitriou

Lifespan Learning And Sleep Laboratory UCL, Institute of Education

Abstract

Sleep is incredibly important for healthy physical, behavioural, and cognitive development throughout childhood. In a world of 24-hour distractions, sleep research with children is of the utmost importance. In this chapter, we consider the methodological tools available to the paediatric sleep researcher, as well as some practical considerations associated with the use of each of these with developmental samples. We then discuss how sleep varies amongst atypical developmental populations, and how to employ each tool in order to obtain optimal data, whilst remaining sensitive to the specific sets of difficulties associated with atypical populations.

13.1 Introduction: Sleep research with children

In a world of increasing environmental distractors, the appeal of getting a good night's sleep is diminishing for many children and adolescents. And yet, more and more research is surfacing showing sleep to be fundamental for healthy development. Firstly, sleep promotes the production of growth hormones, which facilitate healthy brain and bodily development. Secondly, sleep supports processes of memory consolidation that lead to stable long-term memories, and facilitate learning. Finally, sleep helps to support healthy brain development, by promoting neural plasticity and ridding the brain of the neurochemical toxins that build up during the day (Xie et al., 2013). Similarly, we now know that insufficient sleep duration and poor sleep quality have negative effects on academic performance and daytime behaviour. Children with poor sleep perform worse on schoolwork, find it harder to concentrate, and display more disruptive behaviours (Sadeh, Gruber, & Raviv, 2002).

Given that physical growth, knowledge acquisition, and brain development are all at the centre of healthy childhood development, understanding sleep through comprehensive research is vital. There are a variety of research tools that allow us to examine sleep in children, ranging from simply asking about sleep behaviours to measuring the neural or biological activity underlying sleep. Each method comes with its distinct set of benefits and costs, which will be considered further as we work through a range of instruments available. The paediatric sleep researcher should bear in mind that owing to the distinct benefits and costs associated with each technique, using a combined approach is usually advocated.

13.2 Overview of sleep research tools

Sleep poses a difficult problem for the researcher: How can we ask a person to help us understand an activity for which they themselves are not conscious? When talking to a participant about their sleep, you are asking them to give their perspective on an activity about which they are only vaguely aware. Researching sleep in children further compounds this issue, as a child's own insight into their sleep may be incomplete or not easy to glean. In many cases, this means asking the participant's primary caregiver (using questionnaires or interviews) or relying on objective physiological and biological measures, which monitor

sleep patterns through brain activity (polysomnography), movement (actigraphy), or hormone secretion.

This section will first take you through the variety of participant response measures of sleep, and then go on to describe physiological and biological measures. We will compare and contrast the types of information that each tool can be used to obtain, whilst also considering their relative strengths and weaknesses. This information is crucial in selecting the appropriate tool to allow the researcher to capture the most relevant information for the specific purposes of their research. We will pay particular attention to the balancing act between objective reliability and relative disruption to habitual sleep patterns.

13.2.1 Participant response measures: Questionnaires, sleep diaries, and interviews

Participant response measures are assessment tools that question the participant or primary caregiver about their sleep. Currently, the most popular measures are sleep diaries/logs, questionnaires, and interviews. They allow for the capture of abundant descriptive data, which can either be quantitative or qualitative in nature. The researcher is able to extract information that is directly relevant to their research question by adapting existing assessment tools, or developing their own. Such tools represent a relatively inexpensive and non-invasive means of collecting sleep data, making them accessible to researchers without major funding. The aim of each participant response tool is to obtain useful insight into the participant's nighttime behaviour. However, each will vary slightly in terms of the type of information it is able to capture. It is important to understand the distinctions in order to select the most appropriate instrument for your study's needs.

One key distinction is whether you are interested in habitual sleep patterns (how an individual typically sleeps), or immediate sleep patterns over a set period of time (how they sleep over the course of time). Because of the immediate nature of sleep diaries or logs, these will chiefly monitor the latter. *Sleep diaries or logs*, include a selection of questions about sleep behaviours to be answered on a daily basis over a set period of time. For

example, they can provide insight into bed- and rise-times, the number of nighttime awakenings, the types of nighttime disturbances (such as nightmares or toilet trips), and concurrent daytime mood fluctuations. They are commonly used when trying to associate sleep with a measurable daytime activity over the same period of time. For example, one could look at the direct relationship between exercise and sleep, by measuring or manipulating daily exercise and recording concurrent sleep behaviours using a sleep diary. They have the advantage of being easily adapted so that the child themselves can complete them (under adult supervision to avoid errors).

Questionnaires have the benefit of being able to capture either immediate or habitual sleep data. Generally speaking, questionnaires collect more information at one time than sleep diaries. For this reason they commonly require the parent's contribution. Long questionnaires are deemed too demanding for young children, and older children and adolescents with intellectual impairments.

Finally, *interviews* can be used to gain deeper insight into a child's sleep behaviours and perceptions. Like questionnaires, interviews can be used to collect immediate or habitual sleep data. On the whole, interviews obtain rich qualitative data from questions that can be modified to fit the study objectives exactly. They have the additional benefit that the researcher can flexibly discuss new constructs as they arise through conversation. Interviews can be performed with the child, the primary caregiver or both. However, this is a skill that is not to be taken for granted. The ability to listen and guide participants into discussing topics of interest without interrupting the natural flow is not easy. Before undertaking interview research, the researcher should ideally be trained in interview skills with children (see Chapter 6 for a detailed discussion of interviews with children).

Another key distinction is whether you wish to produce a quantitative, a qualitative, or a mixed methods report. All three of the instruments above can collect either quantitative or qualitative data. The key to knowing which you wish to produce, is whether you wish to

describe sleep patterns in a few individual cases in depth, or whether you wish to investigate sleep in a larger sample (using statistical analyses) with the aim of generalising to a wider population. The researcher can ascertain whether an existing participant response tool is qualitative or quantitative by looking at the questions that are asked. Although there are exceptions to any rule, typically closed questions produce quantitative data; data that will be coded into numbers and statistically analysed. *Closed questions* can take the form of dichotomy or binary items (e.g., Did you go to bed at a normal hour? yes/no), multiple category items (e.g., Circle the mood that best describes how you have felt today; happy/grumpy/sleepy/alert) or continuum or scale items (e.g., How did you sleep? very well/ well/ average/ not well/ not well at all). Alternatively, *open questions* produce qualitative data, comprising unrestricted descriptions of sleep-related behaviours, attitudes, and beliefs. The data derived from open questions are less constrained, and therefore reflect the respondent's personal opinions more closely. For example, if I asked you to tell me what mood describes best how you have felt today, you may pick one of the four items suggested above in the multiple category items. However, you may have felt differently from the moods described in the list, or indeed felt a broad description of moods throughout the day. The open-ended interview format is aimed at eliciting a freer response. To this ends, as stated above, it necessitates adequate training in order to usefully engage with the interviewee and be responsive to emerging themes of interest.

Finally, the type of information acquired depends on the respondent. In paediatric sleep research, most of the information is gathered from the child's primary caregiver. One can reasonably assume that the parent of a young child will be able to provide more detailed information, to a greater degree of accuracy, and with fewer misunderstandings, than the child themselves. That said, it is still extremely important that questions are succinct and unambiguous in order to reduce misunderstanding or misinterpretation. As a rule of thumb, questionnaires should take less than 20 minutes to complete (Spruyt & Gozal, 2011), so as to avoid response fatigue or carelessness. The researcher may be interested in the views and perceptions of the child about their own sleep, in which case it is of utmost importance to select a participant response tool that is age-appropriate. The instrument must match the level of understanding of the respondent and be in an easily accessible, age-appropriate

language. The process of designing and validating interviews or questionnaires is a lengthy one. Spruyt and Gozal (2011) give a good explanation of the processes and problems associated with doing so. It is advisable for the novice, or even intermediate researcher, to select an established and well-validated research tool. Whilst participant response tools represent a cheap and easy way to collect sleep data about child participants, they have one key drawback, the issue of subjectivity (see Box 13.1).

<<INSERT BOX 13.1. HERE >>

The issue of subjectivity is compounded further in sleep research, in that the responses gained are in fact the participant's experience of the waking periods surrounding sleep. The participant cannot give you an accurate reflection of the sleep period itself, because by definition they are not conscious for it. For example, if I were to ask you how you slept last night, you might tell me you slept well, based on the fact that you did not have any trouble getting off to sleep and that you woke feeling refreshed. You might tell me you slept poorly, because you found it hard to get to sleep and awoke throughout the night. However, your perception of how the participants have slept may be inaccurate in terms of the processes that underlie sleep. During typical sleep, individuals cycle through sleep stages throughout the night. Each sleep stage is associated with a different kind of sleep (with a distinct pattern of brain waves) that is important for different sleep-dependent processes. From a participant response measure, a participant might relay that they had a good night's sleep, however this might not be reflected in the quality of sleep in terms of these underlying processes. For these reasons (considering Box 13.1), it is advisable for the paediatric sleep researcher not to rely on participant response methods alone; they are best accompanied by a more objective measure of sleep.

13.2.2 Physiological measures

13.2.2.1 Polysomnography

Polysomnography (PSG) is the method of measuring sleep by recording the neural activity that occurs during it. It has long since been considered to be the gold standard in sleep

research, because it is considered an accurate and truly objective observation of the processes underlying sleep. It provides the researcher with a wealth of information about when and how a participant has slept, as well as revealing the sleep stages that they went through and how long they spent in each. PSG has helped identify five physiologically distinct sleep stages, which humans typically cycle through throughout the night. Sleep stages 1-4 are classified as non-rapid eye movement (NREM) sleep and each increases in depth. NREM stages 3 and 4 represent slow wave sleep (SWS), and are sometimes considered together as one stage. The fifth sleep stage is identified by rapid eye movements (REM), in which most of the rest of the muscles in the body are paralysed. PSG has advanced sleep research as a whole, in highlighting the importance of each individual sleep stage to specific cognitive processes. For example, from polysomnographic studies we now know that sleep-dependent learning in children is associated with increased SWS (see Hoedlmoser et al., 2014)

PSG records physiological activity from a small number of electrodes placed on the participant's scalp, face, neck and chest. These electrodes record activity across four domains; brain activity (electroencephalography; EEG), eye movements (electro-oculography; EOG), muscle activity (electro-myography; EMG) and heart rhythms (electro-cardiography; ECG). This data is then sent to a central box, via a system of wires, from which it can be downloaded and analysed offline. The EEG electrodes record the underlying electrical impulses produced by neurons. Because the brain produces different patterns of activity during different sleep stages, the recordings can be scored and categorised accordingly. With this information, we are then able to detect whether a child's sleep activity is typical or atypical. One of the key advantages of PSG, over and above any other sleep measure, is that we are also able to measure whether daytime activities or cognitive events influence sleep, increasing or decreasing certain sleep stages.

The key drawback to polysomnography is its potential to disturb sleep. The equipment itself is cumbersome; sensors are applied around the participant's head and chest. Unsurprisingly, this can feel awkward to the participant, and even threatening to a child participant.

Furthermore, owing to the technical equipment, testing typically takes place in a laboratory. This takes the child out of their natural environment. All of which is likely to upset natural sleep. Newer mobile PSG technologies, allow for the data to be collected in the participant's own home, which better accommodates their normal nightly routine. Nonetheless, it is assumed that the unnatural equipment will introduce some disruption. To attenuate this issue, many studies perform one night of adaptation, in order to get the participant used to sleeping with the apparatus on, and then data is collected on the second night.

13.2.2.2 Actigraphy

Actigraphy is the method of measuring sleep/wake cycles through movement. It works on the assumption that participants will be physically active whilst they are awake and inactive during sleep. Whilst traditionally PSG has been considered the gold standard of sleep research, actigraphy has become increasingly popular in academic research. This is because it has a number of key benefits over and above PSG, and yet retains a high level of reliability with it. Some researchers suggest that the agreement between PSG and actigraphy measures in children is around 85-88% (Hyde et al., 2007).

<<FIGURE 13.1 APPROXIMATELY HERE >>

Movement is measured using an accelerometer set into a small device (which typically takes the form of a wrist watch, see Figure 13.1) that the participant wears for a set period of time. The accelerometer measures and stores units of movement sampled several times a second, which can then be downloaded and analysed offline. The output of the actigraph analysis produces a range of sleep variables in terms of individual and averaged nightly activity. Nightly measures include; sleep onset, sleep efficiency, sleep bouts, time in bed, and time asleep, nighttime awakening, time awake during bedtime, amongst other. Averaged measures include: the most active and least active hours (providing both time of onset and average movement), relative amplitude between the most and least active hours, and the inter- and intra-daily stability of movement patterns. All of these measures give us an idea of the entire sleep/wake cycle of each child participant.

As you can see, there is a wide variety of information available to the researcher from actigraphy data. The data available reveals information both about sleep duration and sleep quality. The richness of the data covering actual sleep quality is greater than that which could be collected using participant response instruments, and is not liable to subjectivity. There are also a number of key benefits that make it more appealing to use than PSG: namely, that it is relatively inexpensive, non-invasive, and involves little or no disruption to the participant's habitual sleep routine. The participant is able to move freely during the day and night, which in itself renders actigraphy data more generalisable to typical daily life. Importantly, actigraphy apparatus is less invasive than PSG, making it a more attractive option for use with children, especially those with developmental disorders. Like all methods, it does have associated costs. The method does not measure sleep per se, rather it infers sleep indirectly by measuring movement. The way the watch is attached can also introduce some inter-individual variability. A watch might be more tightly fitted on one individual than another, which will affect the recording but may not be taken into consideration in data analysis.

13.2.3 Biological measures

An alternative method of studying sleep patterns is by means of examining the biological mechanisms that regulate the sleep/wake cycle. The human sleep/wake cycle is controlled by a central internal timekeeper in the brain, called the suprachiasmatic nuclei. This regulates the expression of a group of genes known as CLOCK genes, which in turn prompt the release of the sleep/wake hormones melatonin and cortisol. By monitoring daily fluctuations in cortisol, melatonin, and CLOCK gene expression, we can understand the typical (and atypical) biological patterns that promote efficient sleep. It is important to bear in mind that such measures do not examine sleep per se, but the circadian rhythm that supports sleep. The chief benefit of using biological measures with children is that they are obtained simply by means of buccal (mouth) samples or saliva swabs. This makes them relatively non-invasive and extremely child-friendly.

These biological systems have a consistent and measurably daily pattern that is largely comparable across healthy individuals. The hormones fluctuate in a diurnal pattern, meaning they typically show one peak and one trough in roughly 24 hours. Hence they are known as circadian rhythms, from the Latin *circa*- (meaning roughly) and *-diem* (meaning a day). The hormones cortisol and melatonin work in tandem. Cortisol aids awakening with cortisol levels peaking in the morning and diminishing throughout the day (Buckley & Schatzberg, 2005). Melatonin has the opposite pattern. Its role is to aid sleep. Melatonin is low in the morning and peaks in the evening just before rest (Waldhauer, Kovacs, & Reiter, 1998). CLOCK genes are more complex. Their patterns of expression vary according to the specific gene. Understanding the rhythmicity of these endocrine and molecular measures, has allowed researchers to investigate their role in sleep and neurodevelopmental disorders (for an explanation of how CLOCK genes apply to childhood disorders see Dueck, Thome, & Hassler, 2012).

An important practical issue to consider here is that the time and number of samples the researcher obtains can have a meaningful effect on the results. Some studies focus on one or two key time points. For example, cortisol peaks roughly 30 minutes after waking (known as the cortisol awakening response; CAR). Consequently, many studies have compared this one time-point between experimental groups (e.g., Zinke, Fries, Kiegel, Kirschbaum, & Dettenborn, 2010). Other studies focus on the entire circadian cycle, sampling a number of time-points throughout the day (e.g. Baird, Coogan, Siddiqui, Donev, & Thome, 2012). These two methods are commonly compared between studies, but often produce discrepant results. This is because they each afford the researcher slightly different insight. For example, one time-point measures have lead to confusion about whether ADHD involves higher or lower cortisol levels than normal(e.g. Ma, Chen, Chen, Liu & Wang, 2011; and Wang et al., 2011). However, recent circadian rhythm studies (sampling fluctuations in cortisol over 24 hours) show that the full diurnal pattern is unstable (e.g. Imeraj et al., 2012). This explains the discrepancy in hyper- or hypo-arousal in the ADHD group; rather than being consistently high or low at a given point during the day, cortisol fluctuates in a less consistent manner throughout the day.

Biological measures allow the researcher to weigh up the relative appeal of avoiding disruption to habitual sleep patterns and increasing objective reliability. If maintaining habitual sleep patterns is more important to the study's objectives, endocrine and molecular measures can be taken throughout the day and on either side of sleep only. In this case, typical sleep patterns are not disturbed. However, this method will not provide an objective measure of sleep itself, but only of the waking hours either side of it. If the researcher is interested in how these biological mechanisms fluctuate during sleep, they must take a number of readings throughout the night, waking the participant intermittently. In this case, the researcher increases the objective reliability as a measure of sleep, but with the associated cost of disturbing the participant's sleep cycle. On the whole in sleep research with children, researchers favour the former approach.

13.2.4 Summary of sleep research tools: A combined approach

There are a number of well-established tools available to the paediatric sleep researcher, each with its own unique set of benefits and costs (see Box. 13.2). Participant response measures provide information about approximate sleep duration and typical daily sleep patterns by collecting subjective data about sleep behaviours. Actigraphy offers a means of objective information about sleep quantity and quality by monitoring nocturnal movement in order to indirectly infer sleep patterns. PSG extends further providing direct objective information about sleep quantity, sleep quality, and sleep stages, by monitoring the neural activity associated with sleep. Biological measures provide an observation of the underlying circadian rhythms that prompt the sleep/wake cycle. In order to select the most appropriate tool for your particular study, you must have a clear understanding of the kind of data that will best help answer your research questions. Perhaps the most important consideration is the balance between maintaining habitual sleep behaviours during the course of participation and gaining a truly objective insight into sleep patterns.

<<INSERT BOX 13.2. HERE>>

Whilst each has its individual use in paediatric sleep research, without doubt the most reliable approach to collecting sleep data is to combine one or more sources of information. In this sense, the data derived from one can be used to support that derived from another. For example, actigraphic analyses require bed and rise times to be inputted, in order to establish individual levels of day and night time activity. Hence, it is necessary to compliment actigraphy data with a simple sleep diary or log. Likewise, participants undergoing PSG will usually only do so for one or two nights, and so using a parent-report measure of habitual sleep behaviours is a good way of augmenting one-off PSG data. Baird and colleagues (2012) provide an excellent example of using multiple methods to produce a comprehensive study of sleep.

13.3 Atypical populations

Sleep is a particularly interesting subject in children with neurodevelopmental disorders. Sleep is important for neural plasticity (the way in which the brain functionally develops according to new knowledge and experience). Sleep allows for the strengthening of existing neural connections and the formation of new ones. As such, disturbed sleep interrupts this important process and may therefore have long-lasting functional consequences on the brain, and general daily performance. Given that sleep problems are prevalent in a number of neurodevelopmental disorders, and that severity of sleep disturbances can contribute to the manifestation of such disorders, research has turned to the investigation of sleep in trying to understand and explain some of these issues. In this section, we will briefly look at some of the differences in sleep that have been reported across three neurodevelopmental disorders, with a focus on the research methods that have allowed such insights. We will also contemplate some practical considerations for using sleep measures with atypical populations.

13.3.1 Autism Spectrum Disorder

Autism Spectrum Disorder (ASD) is a well-known neurodevelopmental disorder affecting three core components: social interactions, communication, and stereotyped repetitive behaviours (Le Couteur et al., 1989). In the UK in 2010, prevalence rates were estimated at roughly 1/1000 for girls and 4/1000 for boys (Taylor, Jick, & MacLaughlin, 2013). Disorders

of sleep co-occur in 50-85% of children with ASD (Richdale, 1999; Xue, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner, 2008). One reason for the sizeable range in reported co-occurrence could be due to the methods used to obtain the estimates. Parental reports consistently describe atypical sleep onset delay (an increased amount of time between going to bed and falling asleep), nighttime awakenings and parasomnias (atypical movements or behaviours during sleep, e.g. sleep walking). However, whilst some parents might be very aware of their child's sleep difficulties, others may not. Indeed, some specialists suggest that children with ASD who suffer from sleep disturbances, rarely complain about their issues to their parents. A recent polysomnographic study (Limoges, Bolduc, Berthiaume, Mottron, & Godbout, 2013) has confirmed differences in sleep onset (delayed), sleep efficiency (reduced), and percentage of time spent awake following sleep onset (increased). The authors further reported increased percentage of sleep stage 1 (NREM) and decreased slow wave sleep (SWS). Interestingly, the same study found no difference in total sleep time. These results show that there may be underlying differences in sleep that would not have been identified if PSG had not been performed. In a population that is not prone to complain about their difficulties, sleep disturbances may prevail without parents being aware of them.

13.3.2 Williams Syndrome (WS)

WS is a neurodevelopmental disorder caused by deletion of some 28 genes on the long arm of chromosome 7q11.23, with around a 1/20,000 prevalence rate (Schubert, 2009; Tassabehji, 2003). It is characterized by overly social behaviour and good expressive language, despite having an IQ range between 50-70. Again, sleep disturbances are often reported. Parents report problems settling, night awakenings, bedtime resistance, and daytime sleepiness (Annaz, Hill, Ashworth, Holley, & Karmiloff-Smith, 2011). A recent actigraphy study (Ashworth, Hill, Karmiloff-Smith, & Dimitriou, 2013) confirmed the presence of longer sleep onset latencies in children with WS compared to controls. However, despite parental reports, the authors found no difference in sleep duration or the number of night awakenings. There are two practical considerations with regards to these discrepant findings. Firstly, children with WS have difficulty self-soothing (getting themselves back to sleep of their own accord), and are therefore more likely to alert their

parents. This means that parents of children with WS may be more aware of nighttime awakenings than those of TD controls. Parent report measures are likely to reflect this. Secondly, it may be that parents of children with WS are more sensitive to their child's daily behaviours, being more responsive to minor difficulties than parents of TD children may be. In this sense, if the child wakes during the night the parent might be more likely to a) be aware of it, and b) view it as an issue.

Polysomnographic studies have shown reduced REM, reduced NREM sleep stages 1 and 2, and increased SWS in children with WS. This pattern is almost the exact reverse to that of ASD. This highlights a key practical issue with both participant response measures and actigraphy; on the surface, sleep disturbances may present in a similar way, yet the underlying mechanisms may be completely different. So whilst parent reports and actigraphy are useful in identifying and highlighting problematic sleep patterns, only PSG is able to allow us insight into the mechanisms underlying such differences (Mason et al., 2011).

13.3.3 *Attention Deficit Hyperactivity Disorder (ADHD)*

ADHD is characterised by the core behavioural symptoms of inattention, hyperactivity, and impulsivity. In the UK, ADHD prevalence rates vary between 1.7-3.8% in school-aged boys, with the prevalence in girls being much lower (around 0.8%; Russell, Rodgers, Ukoumunne, & Ford, 2014). Sleep disturbances are commonly reported in ADHD. Indeed, many researchers have reported that sleep problems correlate strongly with the behavioural manifestation of ADHD (Choi, Yoon, Kim, Chung, & Yoo, 2010; Gruber et al., 2011). However, despite numerous parental reports of sleep disturbances in ADHD, actigraphy and PSG studies have produced inconsistent findings. Whilst the findings of some actigraphy studies are consistent with parent reports (Moreau, Rouleau, & Morin, 2014), others are not (Hvolby, Jorgensen, & Bilenberg, 2008). PSG data too have been inconsistent. Prehn-Kristensen and colleagues report three studies, in which one shows increased REM sleep (confirmed by Kirov et al., 2004) and shorter SWS latency in ADHD children (Prehn-Kristensen et al., 2011a), and two others that find no differences between ADHD

participants and controls (Prehn-Kristensen et al., 2011b; 2013). Owing to discrepancies in PSG data, researchers have begun investigating neural activity during sleep using more sensitive, high-density EEG arrays. Such studies have found immature SWS distribution through the cortex (Ringli et al., 2013) and differences in arousal activity (Miano et al., 2006). In a study of the biological mechanisms underlying sleep, Baird et al. (2012) report that the typical circadian expression of CLOCK genes is lost in the ADHD sample. These studies highlight the seemingly inconsistent results that different methodologies in sleep research can produce. Relying on one measure may bias the results towards the information that measure is able to trace. However, this may not be the full story. So whilst some actigraphy and PSG studies might not pick up on a difference in sleep between ADHD and control, more sensitive methods might.

<<INSERT BOX 13.3. HERE >>

13.4 Selecting a tool: Questions to ask yourself

As we have seen, there is a variety of sleep research tools available for use with children. The paediatric sleep researcher must be aware of their relative strengths and weaknesses, and consider them with respect to their own specific research questions. Firstly, consider your research objectives: Are you interested in habitual sleep patterns, or immediate sleep over the course of participation? Do you want to produce a qualitative or quantitative report? Are you interested in the subjective experience of sleep, or the objective observation of it? Are you interested in sleep patterns, sleep stages, or circadian rhythms? Secondly, you must consider your sample and their capacities: How old is your sample? Do they have any specific difficulties that might influence your choice of tool? If you are interested in the subjective experience of sleep, do they have the capacity to give you adequate information on their sleep habits themselves, or will you rely on parent-report? Are you confident and trained in performing interviews with children? If you are interested in an objective observation of sleep, will the child be comfortable with the equipment? How might the equipment affect their typical sleep patterns? Does this represent a meaningful problem for your report? Finally, each researcher has a different set of resources available to them. It is important to consider your means: How long will testing take per participant?

Do you have access to the preferred assessments or equipment? Do you have the time and the funding to perform a study with physiological or biological measures? Will a participant response measure allow you to capture sufficient information to answer your research question?

If you consider all of these questions with respect to the information covered in this chapter, you should be in a good position to select a tool to fit your research objectives. Remember, that the answers to these questions need not be either/or. Indeed, a mixed methods approach will allow you to obtain a more holistic perspective of sleep (see section 13.2.4). If you have selected a parent-response measure, would a simple participant response measure with the child help corroborate the caregiver's perceptions? If you have decided to investigate immediate sleep over the course of participation, a participant response measure of habitual sleep will enable you to see if this sleep is typical. If you have selected a physiological or biological measure, a sleep diary will help validate the sleep/wake information a physiological measure returns. Ultimately, before embarking on sleep research with children, it is important to think fully about the information you wish to examine, and the most reliable ways of obtaining it with your given sample.

13.5 Summary

Sleep research in children is both interesting and important. The way sleep develops across childhood, how sleep influences daytime learning and behaviour, and the effects of disturbed sleep are all worthy topics of research. There are a variety of tools available to the paediatric sleep researcher, each providing slightly different information, and each with its unique set of strengths and weaknesses. Participant response measures provide an abundance of data with minimal disruption to sleep. However, they are victim to subjective bias. On the whole, physiological methods represent reliable, objective measures of sleep. They vary in the objective information they provide, and their relative disruption to sleep. PSG allows an in depth objective observation of sleep in terms of quality, quantity, and sleep stages. However, it is expensive and fairly invasive for use with children. Actigraphy is non-invasive and much less expensive, but the information it provides is indirect (measuring

movement rather than sleep itself) and comparatively less detailed. Biological measures represent a method of obtaining objective information about circadian rhythms, and are easy to use with children. However, such methods do not measure sleep patterns per se. Instead, they provide insight into the mechanisms underlying the daily sleep/wake cycle. The choice of sleep tool(s) selected will ultimately affect the capacity the paediatric sleep researcher has to answer their research question. For this reason, these methodological considerations must be well researched before testing begins.

Practical tips for paediatric sleep research

- Know your sample, and their capabilities: This will inform your selection of tool and study design.
- Select your tool on the basis of your research question and the data you wish to capture.
- Use a combined approach to data collection: This will allow a more rounded perspective of sleep.

References

Annaz, D., Hill, C. M., Ashworth, A., Holley, S., & Karmiloff-Smith, A. (2011). Characterisation of sleep problems in children with Williams syndrome. *Research in Developmental Disabilities, 32*, 164–169.

Ashworth, A., Hill, C. M., Karmiloff-Smith, A., & Dimitriou, D. (2013). Cross syndrome comparison of sleep problems in children with Down Syndrome and Williams Syndrome. *Research in Developmental Disabilities, 34*, 1572-1580.

Baird, A. L., Coogan, A. N., Siddiqui, A., Donev, R. M., & Thome, J. (2012). Adult attention-deficit hyperactivity disorder is associated with alterations in circadian rhythms at the behavioural, endocrine and molecular levels. *Molecular Psychiatry*, *17*, 988-995.

Buckley, T. M., & Schatzberg, A. F. (2005). On the interactions of the hypothalamic- pituitary adrenal (HPA) axis and sleep: Normal HPA axis activity and circadian rhythm, exemplary sleep disorders. *Journal of Clinical Endocrinology & Metabolism*, *90*, 3106-3114.

Choi, J., Yoon, I. Y., Kim, H. W., Chung, S., & Yoo, H. J. (2010). Differences between objective and subjective sleep measures in children with attention deficit hyperactivity disorder. *Journal of Clinical Sleep Medicine*, *6*, 589–595.

Dueck, A., Thome, J., & Haessler, F. (2012). The role of sleep problems and circadian clock genes in childhood psychiatric disorders. *Journal of Neural Transmission*, *119*, 1097-1104.

Gruber, R., Wiebe, S., Montecalvo, L., Brunetti, B., Amsel, R., & Carrier, J. (2011). Impact of sleep restriction on neurobehavioral functioning of children with attention deficit hyperactivity disorder. *Sleep*, *34*, 315–323.

Hoedlmoser, K., Heib, D. P., Roell, J., Peigneux, P., Sadeh, A., Gruber, G., & Schabus, M. (2014). Slow sleep spindle activity, declarative memory and general cognitive abilities in children. *Sleep*, *37*(9), 1501-1512.

Hvolby, A., Jorgensen, J., & Bilenberg, N. (2008). Actigraphic and parental reports of sleep difficulties in children with attention-deficit/hyperactivity disorder. *Archives of Pediatrics and Adolescent Medicine*, *162*, 323–329.

Hyde, M., O'Driscoll, D. M., Binette, S., Galang, C., Tan, S. K., Verginis, N., ... Horne, R. S. (2007). Validation of actigraphy for determining sleep and wake in children with sleep disordered breathing. *Journal of Sleep Research*, *16*(2), 213-216.

Imeraj, L., Antrop, I., Roeyers, H., Swanson, J., Deschepper, E., Bal, S. & Deboutte, D. (2012). Time-of-day effects in arousal: disrupted diurnal cortisol profiles in children with ADHD. *Journal of Child Psychology and Psychiatry*, *53*, 782–789.

Kirov, R., Kinkelbur, J., Heipke, S., Kostanecka-Endress, T., Westhoff, M., Cohrs, S., ... Rothenberger, A. (2004). Is there a specific polysomnographic sleep pattern in children with attention deficit/hyperactivity disorder? *Journal of Sleep Research*, *13*, 87-93.

Le Couteur, A., Rutter, M., Lord, C., Rios, P., Robertson, S., Holdgrafer, M., & McLennan, J. (1989). Autism diagnostic interview: A standardized investigator-based instrument. *Journal of Autism & Developmental Disorders*, *19*, 363–387.

Limoges, E., Bolduc, C., Berthiaume, C., Mottron, L., & Godbout, R. (2013). Relationship between poor sleep and daytime cognitive performance in young adults with autism. *Research in Developmental Disabilities*, *34*, 1322–1335.

Ma, L., Chen, Y. H., Chen, H., Liu, Y. Y., & Wang, Y. X. (2011). The function of hypothalamusepituitary adrenal axis in children with ADHD. *Brain Research*, *1368*, 159-62.

Mason, T. B., Arens, R., Sharman, J., Bintliff-Janisak, B., Schultz, B., Walters, A. S., ... Pack, A. I. (2011). Sleep in children with Williams Syndrome. *Sleep Medicine*, *12*, 892–897.

Miano, S., Donfrancesco, R., Bruni, O., Ferri, R., Galiffa, S., Pagani, J., ... Pia Villa, M. (2006). NREM sleep instability is reduced in children with attention-deficit/hyperactivity disorder. *Sleep*, *29*, 797–803.

Moreau, V., Rouleau, N., & Morin, C. M. (2014). Sleep of children with attention deficit hyperactivity disorder: Actigraphic and parental reports. *Behavioral Sleep Medicine*, *12*, 69-83.

National Institute for Health and Clinical Excellence (2008). *Attention deficit hyperactivity disorder: Diagnosis and management of ADHD in children, young people and adults*. CG72. London: National Institute for Health and Clinical Excellence.

Prehn-Kristensen, A., Göder, R., Fischer, J., Wilhelm, I., Seeck-Hirschner, M., Aldenhoff, J., & Baving, L. (2011a). Reduced sleep-associated consolidation of declarative memory in attention-deficit/hyperactivity disorder. *Sleep Medicine, 12*(7), 672-679.

Prehn-Kristensen, A., Krauel, K., Hinrichs, H., Fischer, J., Malecki, U., Schuetze, H., ... Baving, L. (2011b). Methylphenidate does not improve interference control during a working memory task in young patients with attention-deficit hyperactivity disorder. *Brain Research, 1388*, 56-68.

Prehn-Kristensen, A., Munz, M., Molzow, I., Wilhelm, I., Wiesner, C. D., & Baving, L. (2013). Sleep promotes consolidation of emotional memory in healthy children but not in children with attention-deficit hyperactivity disorder. *PloS One, 8*(5), e65098.

Richdale, A. L. (1999). Sleep problems in autism: Prevalence, cause, and intervention. *Developmental Medicine & Child Neurology, 41*, 60-66.

Ringli, M., Souissi, S., Kurth, S., Brandeis, D., Jenni, O. G., & Huber, R. (2013). Topography of sleep slow wave activity in children with attention-deficit/hyperactivity disorder. *Cortex, 49*, 340-347.

Russell, G., Rodgers, L. R., Ukoumunne, O. C., & Ford, T. (2014). Prevalence of parent-reported ASD and ADHD in the UK: findings from the Millennium Cohort Study. *Journal of Autism & Developmental Disorders, 44*, 31-40.

Sadeh, A., Gruber, R., & Raviv, A. (2002). Sleep, neurobehavioral functioning, and behavior problems in school-age children. *Child Development, 73*, 405-417.

Schubert, C. (2009). The genomic basis of the Williams–Beuren syndrome. *Cellular and Molecular Life Science*, *66*, 1178–1197.

Spruyt, K., & Gozal, D. (2011). Development of paediatric sleep questionnaires as diagnostic or epidemiological tools: a brief review of dos and don'ts. *Sleep Medicine Reviews*, *15*, 7–17.

Tassabehji, M. (2003). Williams–Beuren syndrome: A challenge for genotype–phenotype correlations. *Human Molecular Genetics*, *15*, 229–237.

Taylor, B., Jick, H., & MacLaughlin, D. (2013). Prevalence and incidence rates of autism in the UK: time trend from 2004–2010 in children aged 8 years. *British Medical Journal open*, *3*(10), e003219.

Waldhauer, F., Kovacs, J., & Reiter, E. (1998). Age-related changes in melatonin levels in humans and its potential consequences for sleep disorders. *Experimental Gerontology*, *33*, 759-772.

Wang, L. J., Huang, Y. S., Hsiao, C. C., Chiang, Y. L., Wu, C. C., Shang, Z. Y., & Chen, C. K. (2011). Salivary dehydroepiandrosterone, but not cortisol, is associated with attention deficit hyper- activity disorder. *The World Journal of Biological Psychiatry*, *12*, 99-109.

Xie, L., Kang, H., Xu, Q., Chen, M. J., Liao, Y., Thiyagarajan, M., ... Nedergaard, M. (2013). Sleep drives metabolite clearance from the adult brain. *Science*, *342*(6156), 373-377.

Xue, M., Brimacombe, M., Chaaban, J., Zimmerman-Bier, B., & Wagner, G. C. (2008). Autism spectrum disorders: Concurrent clinical disorders. *Journal of Child Neurology*, *23*, 6–13.

Zinke, K., Fries, E., Kliegel, M., Kirschbaum, C., & Dettenborn, L. (2010). Children with high-functioning autism show a normal cortisol awakening response (CAR). *Psychoneuroendocrinology*, *35*, 1578-1582.