THE RELATIONSHIP BETWEEN SLEEP AND DAYTIME BEHAVIOUR IN CHILDREN WITH AUTISM SPECTRUM DISORDER

by

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Thesis Overview

This thesis presents the work completed over the course of the author's doctorate of clinical psychology, and consists of two volumes. The first volume includes three chapters, the first of which is a systematic literature review and meta-analysis of the prevalence of insomnia in clinical and non-clinical populations of children and adolescents. The second chapter is an empirical paper exploring the relationship between sleep problems and daytime challenging behaviour in children with Autism Spectrum Disorders. The final chapter is a document summarising chapters one and two for the purpose of public dissemination.

The second volume constitutes each of the five clinical practice reports (CPR's) completed over the course of the doctoral training. The first CPR describes a clinical case from an older adult placement, and compares formulations from two psychological perspectives that could be used to inform intervention with the client. The second CPR describes a single-case experimental design methodology used to determine the effectiveness of a therapeutic intervention with a client presenting with behaviour that care-staff considered challenging. The third CPR consists of a service evaluation conducted to appraise the effectiveness of the referral pathway into a secondary care psychology service, using both quantitative and qualitative methodology. The fourth CPR offers personal reflections on the process of providing a consultancy service to a care provider external to the National Health Service, and finally, the fifth CPR summarises a clinical case study from a placement with a child and adolescent mental health service.

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CHAPTER ONE

The prevalence of insomnia in children and adolescents: A systematic review and meta-analysis

Abstract

Background. Insomnia is characterised by difficulty initiating sleep (DIS), difficulty maintaining sleep (DMS) and early morning awakening (EMA), resulting in distress or impairment in daytime functioning. The prevalence of insomnia in childhood and adolescence has been estimated between 4% and 41% and is associated with physical and mental health conditions. The aim of this research was to synthesise the existing prevalence data on insomnia in childhood and adolescence, adjusting for methodological quality, and to compare this with prevalence estimates observed in clinical samples.

Methodology. One thousand and eighty-nine articles were identified through literature searches. After removal of duplicate articles, and those not meeting eligibility criteria, 92 articles remained and were entered into a quality framework, to weight articles by methodological quality. Ten articles reported data from the same sample as another article in the collection, and were therefore removed, leaving 82 articles to be included in the meta-analyses. Pooled prevalence estimates of insomnia were generated, and compared between the varying definitions of insomnia, and between clinical and non-clinical populations.

Results. Using a quality-effects model, pooled prevalence estimates were generated for insomnia (19.6%), insomnia with daytime impairment (9.9%), insomnia without comorbidities (4.1%), and DIS (13.6%), DMS (9.4%) and EMA (5.6%) in children and adolescents. The prevalence of insomnia in anxiety disorders was 59.0% (Odds Ratio compared to non-clinical population 5.8), 58.7% in depression (OR 5.9), and 54.5% in chronic pain (OR 4.8).

Discussion. This is the first meta-analysis providing synthesised prevalence data for insomnia in the child and adolescent population, across definitions of insomnia. The findings also demonstrate that children and adolescents with comorbid conditions are more likely to present with insomnia. The findings indicate that the definition and methodology used to assess for insomnia should be considered when

interpreting the findings of empirical articles. A balance should be sought between being over-inclusive and conservative when developing criteria, particularly with regard to the exclusion of participants presenting with comorbidities, or who present with sleep problems but without daytime impairment. This limits the extent to which empirical findings can be generalised to clinical practice.

1.1 Introduction

Sleep plays a crucial role in human development and functioning, with disordered sleep having wide-ranging negative consequences for a variety of functions including cognitive fatigue, emotional affect, and motor co-ordination (Banks & Dinges, 2007; Durmer & Dinges, 2005; Pilcher & Huffcutt, 1996; Wickens, Hutchin, Laux & Sebok, 2015). One category of sleep disorder is insomnia. According to the Diagnostic and Statistical Manual: Fifth edition (DSM-V; American Psychiatric Association, 2013) and the International Classification for Sleep Disorders: Third edition (ICSD-3; American Academy of Sleep Medicine, 2014), insomnia is characterised by difficulty falling to sleep, not being able to remain asleep throughout the night, or waking earlier than would be preferred. One or more of these three symptoms must occur at least three times a week, and be present for a minimum of 3 months for insomnia to be diagnosed. Sleep problems must also be present despite there being sufficient opportunity and an appropriate sleeping environment, as well as observable impairments to daytime functioning, in order for a diagnosis of insomnia to be given.

Insomnia has commonly been subcategorised as either ‘primary’, and therefore idiopathic in origin, or ‘secondary’, whereby the insomnia is comorbid with another disorder. Recently, it has been debated whether the distinction between the primary and secondary subtypes of insomnia are as easily defined as the psychiatric taxonomy would imply (Lichstein, 2006). It can be difficult to ascertain the direction of causality, and whether a primary diagnosis is causing the comorbid secondary insomnia, whether insomnia is primary and driving the secondary diagnosis, or whether the two disorders are inter-related, each influencing the severity of the

other. As a result, both the DSM-V and ICSD-3 have combined all subtypes of insomnia under the single term of ‘insomnia disorders’ (Sateia, 2014), and distinguish this only from ‘short-term insomnia disorder’, which refers to insomnia in response to transient factors, such as stress or changes to the environment. This has implications not only within the empirical literature, in terms of whether research samples will now include participants with both primary and secondary forms of insomnia, but also the way in which insomnia is defined for clinical assessment and treatment, as the intervention for insomnia secondary to a comorbid disorder might differ to that offered for a primary insomnia.

Irrespective of the difficulty in determining the direction of causality between insomnia and comorbid conditions, insomnia has been consistently reported as more prevalent in individuals with mental health problems, such as anxiety and depression (Ohayon & Roth, 2003; Taylor, Lichstein, Durrence, Reidel & Bush, 2005). In a large cross-sectional survey of an adult Norwegian population, Sivertsen and colleagues reported significant associations between insomnia and mental health problems, with odds ratios of 1.99 and 2.42 for insomnia in depression and anxiety respectively (Sivertsen, Krokstad, Øverland & Mykletun, 2009). Several cognitive and cognitive-behavioural approaches to treatment for insomnia have been successfully developed as a result of these data (Harvey, 2002; Morin et al., 2006). Furthermore, longitudinal studies have suggested that in some cases insomnia may occur prior to the development of some mental health problems, or persist after recovery, and therefore should not simply be considered a ‘symptom’ of mental health problems which will cease once comorbid conditions subside (Baglioni et al., 2011; Breslau, Roth, Rosenthal & Andreski, 1996; Eaton, Badawi &

Melton, 1995; Johnson, Roth & Breslau, 2006a; Ohayon & Roth, 2003; Weissman, Greenwald, Niño-Murcia & Dement, 1997).

In addition to being more prevalent in individuals experiencing mental health difficulties, insomnia is also observed more frequently in those with a physical health diagnosis than in the general population. These diagnoses range from enduring and highly pervasive conditions, such as cancer or chronic pain, to migraine or allergies (Bennett et al., 2010; Savard & Morin, 2001; Smith, Perlis, Smith, Giles & Carmody, 2000; Sutton, Moldofsky & Badley, 2001). In these instances, the insomnia may be a result of physical discomfort arising from the medical condition, or a side effect of the medication that has been prescribed to the individual. Cognitive or emotional arousal has a negative impact on a person's ability to sleep (Lichstein & Rosenthal, 1980), thus sleep disturbance may be further heightened if an individual's medical condition is causing them distress. Therefore, those with a physical health problem may also experience feelings of anxiety or low mood, which could further influence the likelihood of insomnia being present in this population.

The association between insomnia and physical and mental health conditions has also been well documented in the child and adolescent populations (Desaulniers et al., 2015; Luc, Gupta, Birnberg, Reddick & Kohrman, 2006; Palermo, Wilson, Lewandowski, Toliver-Sokol & Murray, 2011). Researchers have emphasised the importance of identifying insomnia early, because of the likely bi-directional influence on the severity of physical and mental health diagnoses, as well as the long-term impact that sleeplessness can have on a child's development (Jan et al., 2010). It is therefore important that clinicians are aware of the signs of insomnia, and are familiar with the treatment options available. Yet, as Meltzer and

Mindell (2014) highlight, there is no universally-agreed definition of childhood insomnia.

Both the DSM-V and ICSD-3 include childhood insomnia under the overall umbrella of insomnia disorders, rather than listing childhood sleep disorders separately. Other definitions that have been proposed include that by Mindell and colleagues (2006), who suggest that childhood insomnia is the “repeated difficulty with sleep initiation, duration, consolidation, or quality that occurs despite age-appropriate time and opportunity for sleep, and results in daytime functional impairment for the child and/ or family” (page 1225). Glaze, Rosen and Owens (2002), however, suggest that insomnia in childhood should be considered a descriptive term, rather than a diagnostic category. They propose that any given criteria must acknowledge normal changes to sleeping patterns over the course of development, as well as the impact that the family have on the sleep disorder, and that the sleep disorder has on the family. Their proposed criteria are that the child has significant difficulty initiating or maintaining sleep, and that this is perceived as problematic by the child and/ or their parent; the sleeplessness must cause daytime impairment; and is not borne of an alternative sleep disorder, such as narcolepsy or sleep apnoea, or use of substances or medication. The authors acknowledge that the frequency, severity and duration of the difficulty should define the significance of the sleeplessness, but do not suggest what recommended thresholds for these factors might be.

Childhood insomnia is more difficult to define than adult insomnia due to several factors. Firstly, there are natural changes in a child’s sleep trajectory over the course of their development (Blair et al., 2012), and therefore behaviours that

would be considered typical for one age group, may be considered worthy of clinical investigation in another. For example, bedtime resistance and night-wakings are often observed in young children, whereas delayed sleep phase and shortened total sleep duration are more commonly observed in older children and adolescents (Liu, Liu, Owens & Kaplan, 2005; Owens, 2007; Owens, Spirito, McGuinn & Nobile, 2000). Assessment of the sleep problem is also heavily biased by the perception of the parent or care-giver who is often responsible for completion of the sleep diaries or clinical measures. For example, a parent may perceive their child's sleeplessness to be more problematic than does the child, or they may not even be aware of their child's night-time behaviours if they sleep well themselves. A further consideration is that for children and adolescents, sleep and wake times are typically dictated by a parent or care-giver. Where an adult may be able to adjust their bedtime dependent on their level of fatigue or arousal, a child may not be able to do so. Children's night-time behaviours are also heavily influenced by their parents' responses. A child who is allowed to sit downstairs and watch television when they are unable to sleep may have little opportunity or motivation to self-soothe the next time they awaken during the night. All of these factors distort the threshold for when a sleep problem reaches clinical significance and necessitates intervention.

In the research literature, these problems are evident in the wide variety of methodology used to study insomnia in childhood. Several research articles rely on the DSM or ICSD diagnostic criteria to define their construct of insomnia, and base their research items accordingly. It could be argued that these criteria are based upon the presentation of adult insomnia, and therefore are not necessarily applicable to child and adolescent populations. Yet, the definitions developed

specifically for the paediatric population may better capture the nature of childhood insomnia, but are broad and difficult to quantify. Other sleep disorders, such as parasomnias or sleep apnoea could also account for changes to the duration or quality of a child's sleep, as well as impact on daytime functioning, and therefore the definitions may not distinguish childhood insomnia sufficiently from other disorders. Furthermore, the difficulty in identifying possible causality between insomnia and comorbid conditions causes problems for those seeking to conduct research with this group, in terms of trying to recruit a homogenous sample and control for possible confounding variables.

The assessment of insomnia itself can also pose a challenge. For example, it is commonly observed that parent and child reports of the sleep problem differ (Fricke-Oerkermann et al., 2007; Short, Gradisar, Lack, Wright & Chatburn, 2013). As previously mentioned, this may be due to differing perceptions of the parent and child as to the extent of the sleep problem. Furthermore, the existing research literature often does not acknowledge the different components of childhood insomnia when assessing the presence of sleep disorders in the paediatric population. For example, measures do not typically enquire about bedtime routines, the sleeping environment, or the response of parents to the child's sleeplessness. It is therefore difficult to draw conclusions from the literature as to what the clinical picture of childhood insomnia actually looks like. Furthermore, the main symptoms of insomnia are rarely studied in isolation and no synthesised prevalence data exist to the best of the author's knowledge. As a result, it is not possible to determine what the core components of insomnia consist of in this population.

The challenges outlined above make it difficult to study empirically the nature of childhood insomnia, to ascertain the pervasiveness and trajectory of insomnia in the paediatric and adolescent populations, and to synthesise the findings from various research studies. This is problematic not only in terms of understanding the potential burden that insomnia has on clinical resources, but also being able to understand the long-term impact that sleeplessness has on a child's functioning, such as learning, and physical and mental health (Curcio, Ferrara & Gennaro, 2006; Roberts, Roberts & Duong, 2008a). It is also not possible to conclude that populations of individuals with a diagnosed condition present with a higher risk of developing insomnia, without a robust estimate of the prevalence of insomnia in the general population with which to compare. A wide range of prevalence estimates have been reported in the child and adolescent populations, ranging between 4% and 41% (Archbold, Pituch, Panahi & Chervin, 2002; Camhi, Morgan, Pernisco & Quan, 2000). This is highly dependent on the way in which insomnia has been defined, the methodology employed to gauge the presence of insomnia, and the population from which the research sample was drawn.

The present review therefore has several aims:

1. To provide an estimate of the prevalence of insomnia, as well as each of the key symptoms of insomnia, in non-clinical populations of children and adolescents.
2. To provide an estimate of prevalence adjusted for the quality of the research methodology.
3. To compare the prevalence estimates identified for the non-clinical population, with those identified for populations of children and adolescents with physical and mental health conditions.

1.2 Methodology

1.2.1 Search Strategy

The meta-analysis data were gathered and evaluated through the process set out by the PRISMA statement (Moher, Liberati, Tetzlaff, Altman & the PRISMA Group, 2009). This involved identifying the information through an appropriate search strategy, screening the resulting articles for duplicates and eligibility, and finally, processing the remaining articles through a qualitative framework to produce the overall estimates of prevalence.

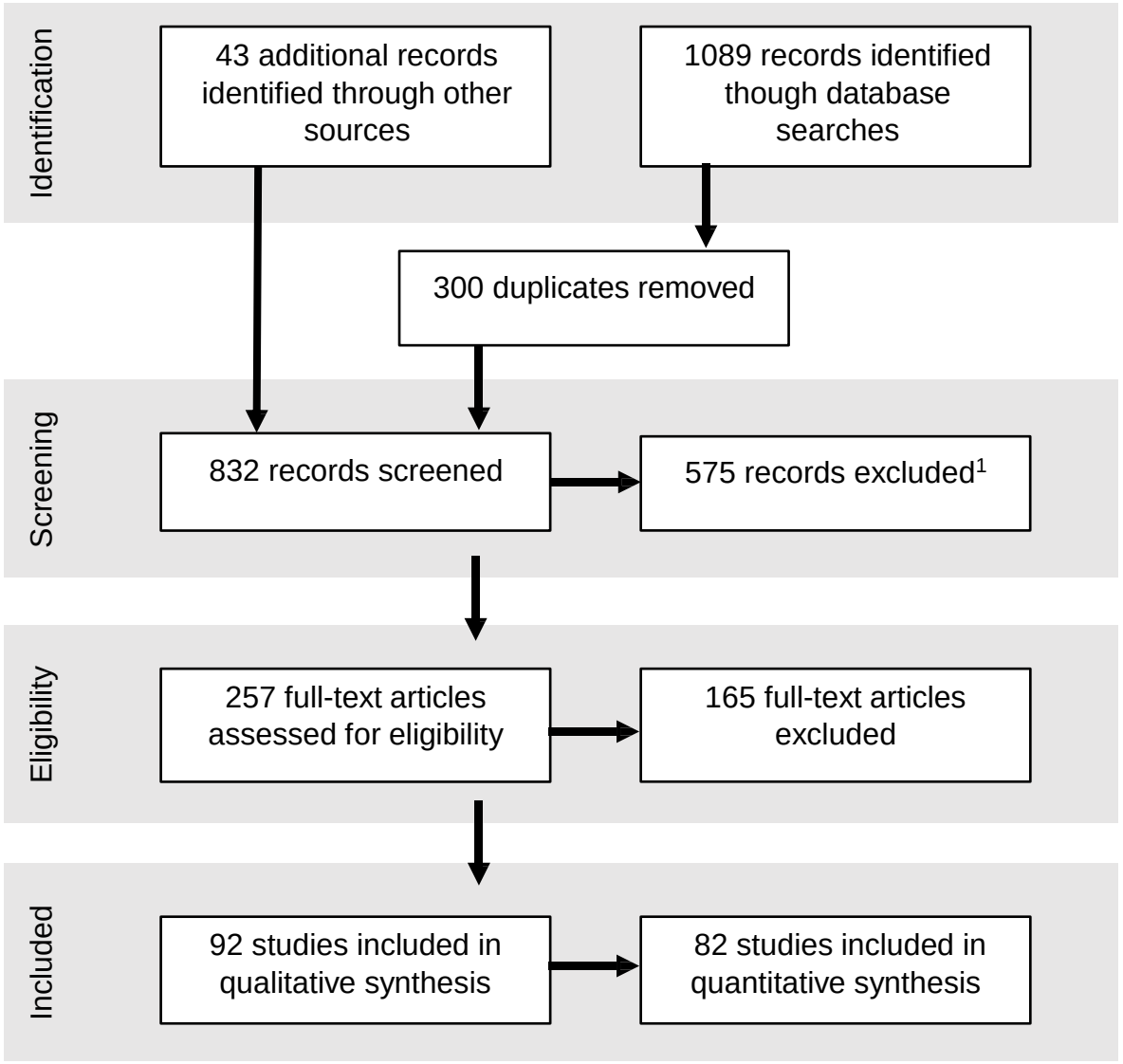

Figure 1.1 - Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement (Moher, Liberati, Tetzlaff, Altman & the PRISMA Group, 2009)

The meta-analysis sought to be as inclusive as possible, and so literature searches were conducted across several databases (Table 1.1). The search terms included terms were 'insomnia' or 'sleeplessness', 'child*,' 'paediatr*,' 'pediatr*,' 'adolesc*,' 'school-age' or 'teenage*' and 'prevalence.' The terms related to insomnia and age of the population were included as a keyword search, and

[1] Inclusive of twenty-nine articles that were not available as full-text.

prevalence was searched for throughout the entire article text. This was to ensure that studies which had reported prevalence estimates alongside other key findings were also included.

Table 1.1 - Databases included in the search strategy

Database	Dates Inclusive	Date Searched
Ovid	1806 – October 2015	16/10/2015
CAB Abstracts		
Embase		
Embase Classic		
HMIC Health Management Information Consortium		
Journals@Ovid		
Ovid MEDLINE® In-Process & Other Non-Indexed Citatons and Ovid MEDLINE®		
Ovid MEDLINE®		
Ovid MEDLINE® In-Process & Other Non-Indexed Citatons		
PsycARTICLES Full Text		
PsycINFO		
Social Policy and Practice		
EBSCO	1977 – October 2015	26/10/2015
AMED		
Child Development and Adolescent Studies		
CINAHL Plus		
ERIC ProQuest		
MEDLINE		
Cochrane Library	1898 – October 2015	26/10/2015
Wiley Online Library	1975 – October 2015	26/10/2015
Web of Knowledge	1975 – October 2015	27/10/2015
Arts and Humanities Citation Index		
BIOSIS Citation Index		
SciELO Citation Index		
Science Citation Index		
Social Sciences Citation Index		

1.2.2 Eligibility screening

A total of 1,089 articles were identified through literature searches, and a further 43 articles were identified through hand-searching of references. Three hundred articles were duplicates. These were removed, leaving 832 articles. The remaining articles were screened and evaluated using the strategies detailed in Tables 1.2-1.4. Each stage of the process comprised evaluating each article against several criteria, ensuring that the remaining articles were relevant to the target population and presented data that were relevant to the research question.

The first stage consisted of screening all articles by title against the criteria listed in Table 1.2. Articles were excluded if they were not available in English, did not present original empirical data, or include a typically-developing sample between the ages of 4-18 years. Papers were also excluded if they were not relevant to the research question. Criteria were applied in the order listed below, such that if an article was a review of insomnia in adulthood, it would be excluded on the basis of not being an empirical study, as this criterion is applied to the article first.

Table 1.2 - Strategy for screening articles by title.

Criteria	Articles to be excluded	Number excluded
English language	Non-English language	46
Empirical study	Reviews, practitioner guides, conference proceedings, meeting abstracts, letter to editors, case studies	39
Typically-developing sample	Sample includes children with a neurodevelopmental disorder	25
Sample of school-age	Sample includes adult or infant populations	157
Relevant	Irrelevant articles, such as pharmacological trials	76
Total		343

The second stage of the screening process was to review the articles by abstract, using the same criteria employed in the initial stage. An additional criterion was included at this stage, whereby the article was excluded if there was a bias in the recruitment strategy described in the abstract. For example, if the sample were recruited based on participants presenting with a related difficulty such as nightmares, or having a risk factor such as diagnoses of insomnia in the family, it was considered that the resulting prevalence estimate would likely be non-representative of the target population, and the article was removed from further analysis.

Table 1.3 - Strategy for screening articles by abstract.

Criteria	Articles to be excluded	Number excluded
Empirical study	Reviews, practitioner guides, conference proceedings, meeting abstracts, letter to editors, case studies	65
Typically-developing sample	Sample includes children with a neurodevelopmental disorder	0
Sample of school-age	Sample includes adult or infant populations	103
Non-biased recruitment	Sample recruited on basis of having a sleep disorder, or disorder related to sleep, such as snoring	13
Relevant	Irrelevant articles where sleep disorder is not focus of study, such as side effect of medication	22
Total		203

Once the articles had been screened by both title and abstract, the remaining papers were subject to full text screening. The criteria applied during the full text screening phase were similar to those in the first two stages. However, articles were also excluded at this stage if they did not report frequency or prevalence statistics. In some cases, this was because descriptive statistics were simply not reported, or

because the data were collapsed with other variables, such as other sleep disorders. During this screening phase, several articles were also removed based on the age criterion. This was because, although the abstract detailed that data for the relevant age group were collected during the study, in the full text screen it was observed that these data could not be separated from that of other age groups. For example, several papers combined data for all participants under 25 years. Consequently, prevalence data for those aged 4-18 years could not be extracted.

Table 1.4 - Strategy for screening articles by full text.

Criteria	Articles to be excluded	Number excluded
Empirical study	Reviews, practitioner guides, conference proceedings, meeting abstracts, letter to editors, case studies	9
Sample of school-age	Sample includes adult or infant populations, or includes target population data but within wider age bracket.	111
Non-biased recruitment	Sample recruited on basis of having a sleep disorder, or disorder related to sleep, such as snoring	3
Relevant	Irrelevant articles where sleep disorder is not focus of study, such as side effect of medication	25
Frequency or prevalence statistics reported	Frequency or prevalence data not reported, or insomnia data combined with other data, such as other sleep disorders.	17
Total		165

Papers that had been identified as meeting the inclusion criteria and that included a clinical sample, such as children and adolescents with a physical or mental health diagnosis, were grouped into a separate collection. This allowed for comparison of prevalence rates of insomnia in clinical and non-clinical samples. Clinical samples where it was likely that there were several comorbid physical and mental health problems or neurodevelopmental disorders, such as genetic syndromes, were not included in the analysis. This decision was made as the

varying influences of each condition on the presence of insomnia could not be ascertained.

1.2.3 Application of the Quality Framework

Following the three stages of the screening process, the remaining 92 articles were evaluated for their methodological quality. Where it was suspected that the same sample had been used as a source of data for more than one article, the article with the highest quality rating was retained, and the others excluded.

The chosen quality criteria focused on sample recruitment and the extent to which the sample was representative of the target population, the authors' definition of insomnia, and the quality and reliability of their assessment of insomnia (Table 1.5). These criteria were based on the framework developed by Munn, Moola, Lisy and Riitano (2014), which is tailored specifically for the evaluation of studies estimating rates of prevalence. As the authors point out, prevalence data can be heavily biased if the sample of a study is not recruited at random or representative of the target population, or if the construct of interest is not accurately measured or well-defined. These factors therefore feature strongly in the chosen quality framework.

Papers that made use of standardised measures, or of multiple forms of methodology to assess for insomnia, were given the highest weighting, as were measures that included items based on the key symptoms of insomnia, or that were explicitly based on diagnostic criteria. Articles in which a single item was used to assess for insomnia, within a measure designed to assess for a construct other than sleep disorders, received a lower weighting. A preferred threshold of 70% participant response uptake was used (Babbie, 2007), with lower rates receiving a

lower weighting. Quality ratings for reported reliability or validity statistics were taken from Landis and Koch (1977), and Streiner and Norman (2002).

Table 1.5 - Quality framework.

Quality rating	Sample recruitment	Response rate	Definition of insomnia	Assessment of insomnia	Assessment validity and reliability
0	Not specified	Not specified <50%	Not specified	Not specified	Not specified k≤ 0.2 α *or* r≤ 0.2
1	Restricted or non-random sample, such as health clinics or a single school	≥50%	Sleeplessness items rated as present/ absent	Parent/ professional opinion	k= 0.21 - 0.4 α *or* r= 0.21 – 0.4
2	Random sample, but with likelihood of response bias	≥60%	Sleeplessness items with indication of frequency or duration of difficulty	Items not described, or measure is not typically used to assess for sleep problems	k= 0.41 – 0.6 α *or* r= 0.41 – 0.7
				Items do not include any of the three main symptoms and are not based on diagnostic criteria	
3	Random or total population sample	≥70%	Based on diagnostic criteria, such as DSM-IV or V, or ICSD	Items include any of the three main symptoms or are explicitly based on diagnostic criteria	k≥ 0.61 α *or* r≥ 0.7
	Sleep data collected as part of wider dataset, reducing likelihood of response bias		Sleeplessness items with indication of frequency and duration of difficulty or mention of daytime impairment	Multiple methods e.g. questionnaire, diagnostic interview, sleep diary, physiological measure	
				Standardised, validated or clinical measure	

By evaluating the articles against the criteria of the quality framework, each was weighted according to its methodological quality. A rating of between zero and three was given for each of the criteria, and the total score was then divided by the maximum total score of fifteen. This gave an overall numerical quality rating for each article of between zero and one, with one indicating a higher quality of paper. If an article received a rating of zero for their recruitment of the sample, their definition of insomnia, or their assessment of insomnia, it was excluded from further analysis.

1.2.4 Analysis

The prevalence estimates reported in each article were identified and pooled using MetaXL 2.0 (Barendregt & Doi, 2011). A random effects model was applied to the data, due to the variability in the reported prevalence rates, the heterogeneity of the sampling and methodology, and the likelihood of sampling error. Use of this model therefore attempted to account for study level differences in addition to sampling error, by incorporating a parameter based on homogeneity of the study data and sampling variance. However, because the random effects model allocates weighting in a manner less dependent on sample size, this can lead to a reduction in the weighting of large high quality studies, and an increased impact of poor quality small studies.

A quality effects model was therefore also included to ensure that pooled prevalence rates also accounted for methodological rigour. To generate the quality effects model, MetaXL uses the sample size in addition to a quality parameter to distribute the total weighting across the studies included in the pooled analysis, such that studies with a larger sample and a greater quality rating receive the highest proportion of available weighting. The quality parameter was generated based on

the method described in section 1.2.3. The inclusion of both models in the present meta-analysis allows the reader to observe the contribution of methodological quality by drawing comparison between the pooled prevalence estimates of the quality- and random-effects models. The meta-analysis therefore allows for extraction of prevalence estimates that account for both the quality of the research, and sources of uncontrolled variability.

In line with the review of the prevalence of insomnia in the adult population published by Ohayon (2002), papers were grouped based on their definitions of insomnia. The first group included any article reporting a prevalence of insomnia, irrespective of the symptoms used to define this term. The following groups included papers that had reported data on a single symptom of insomnia, such as difficulty initiating sleep (DIS), difficulty maintaining sleep (DMS), and early morning awakening (EMA). If a paper reported a prevalence of overall insomnia, but only used a single symptom in their definition and assessment, the article was included under the pooled data for both estimates of the prevalence of overall insomnia, and the symptom alone. Articles that had then included additional criteria, such as the presence of daytime impairment, or insomnia in the absence of other comorbid conditions were grouped separately. Finally, the prevalence of insomnia in physical or mental health conditions were analysed, and odds ratios with 95% confidence intervals were calculated and compared against the overall prevalence estimate for insomnia.

If comparisons were made between two groups in an article, and the two groups were recruited separately in order to obtain comparable sample sizes, only data from the control group were included in the prevalence data. If a population

sample was recruited and then divided according to any naturally-occurring differences, all sample data were included in the prevalence data, as this was considered to be representative of the natural variance occurring within any sample. For example, Zhang et al. (2015) classified a fifth of their sample as overweight, and drew comparisons between participants classified as overweight and the remainder of the sample. Because the participants who were classified as overweight were part of the normal variation of the sample, to only include the control group in the estimates of the prevalence of insomnia, would not be representative of the wider population.

Where prevalence data were not reported, but frequency statistics and sample size were available, prevalence estimates were calculated from these data. In longitudinal studies, prevalence data were only extracted from the time-point with the largest sample, unless it could be concluded that participants could not have been included in the data collection at multiple time-points. The only exception to this was the Pallesen et al. (2008) study, from which data from the most recent time-point were used[1].

1.3 Results

1.3.1 Types of Insomnia in the Non-Clinical Population

Insomnia. The literature search identified 55 papers that reported a prevalence of insomnia within typically-developing children aged 4-18 years. Of these, seven were excluded due to having included data from the same sample as another article (Amaral et al., 2014; Liu, 2004; Liu & Zhou, 2002; Roberts, Roberts & Chan, 2008b; Roberts, Roberts & Chen, 2000; Roberts et al., 2008a; Singareddy

[1] The most recent time-point was chosen, as the largest dataset was 10 years prior to publication.

et al., 2009). A further four papers were removed as they did not describe either their definition or assessment of insomnia, or the manner in which the sample had been recruited (Chen et al., 2011; Chung & Cheung, 2008; Rastogi, Tripathi & Ravishanker, 2010; Yang, Huh, Jeong, Lee & Choi, 2014). This resulted in a total of 44 papers to be included in the pooled meta-analysis. Each of these articles are summarised in Table 1.6. The majority of the articles were published by authors in North America, or countries on the Asian continent, such as Japan or South Korea. There were relatively few articles from Europe or Australia.

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Alvaro et al. (2014)	Multiple schools	2	NS	0	DSM-IV[1]	3	ISI[2]	3	α= .83 r= .79[3]	3 .73	318 (51.6%)	15.0 (1.3) *12-18*	12.5%
Amaral et al. (2013)	Multiple schools	2	82.1%	3	DSM-IV	3	DSM-IV	3	NS	0 .73	6919 (47.0%)	*12-18*	21.4%
Archbold et al. (2002)[4]	Paediatric outpatient clinics	1	52.0%	1	2 of NRS/ DIS/ DMS/ EMA[5]	1	PSQ[6]	3	"good"	2 .53	639 (NS)	*5-14*	17.1%
Barclay et al. (2015)[7]	Longitudinal twin study	1	NS	0	DSM-III-R[1]	3	CAPA[8]	3	NS	0 .47	2789 (46.0%)	Mode= 8.3 *8-17*	19.5%
Blank et al. (2015)	National survey	3	82.9%	3	DSM-IV	3	Interviews based on DSM-IV	3	NS	0 .80	6483 (48.6%)	*13-18*	33.7%
Calhoun et al. (2014)	Population/ stratified school sample	2	70.0%	3	ICSD[9]	3	PBS[10], based on ICSD	3	NS	0 .73	700 (46.0%)	8.8 (1.8) *5-12*	19.3%
Camhi et al. (2000)[11]	Existing sample	3	81.6%	3	DIS/ DMS/ EMA	1	Key symptoms	3	NS	0 .67	314 (55.1%)	*7-14*	17.2%

[1] DSM= Diagnostic and Statistical Manual
[2] ISI= Insomnia Severity Index (Morin, 1993)
[3] α= Cronbach's alpha; r= test-retest reliability/ inter-rater reliability
[4] Over 5 years only
[5] NRS= Non-restorative sleep; DIS= Difficulty initiating sleep; DMS= Difficulty maintaining sleep; EMA= Early morning awakening
[6] PSQ= Pediatric Sleep Questionnaire (Chervin et al., 2000)
[7] Wave 1 only
[8] CAPA= Child and Adolescent Psychiatric Assessment (Angold et al., 1995)
[9] ICSD= International Classification of Sleep Disorders
[10] PBS= Pediatric Behavior Scale (Lindgren & Koeppl, 1987)
[11] 7-14 years only. Although authors do not use the term insomnia, they combine data on DIS and DMS, and so these symptoms cannot be analysed separately.

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Chen et al. (2013)[1]	National survey	3	NS	0	DIS/ DMS/ NRS, often/ always, 4week duration	3	Insomnia self-assess. inventory[2]	3	NS	0 .60	2113 (52.9%)	*15-17*	20.9%
Choi et al. (2009)[3]	Random/ stratified school sample	2	86.7%	3	DIS/ DMS/ EMA, 3+ weekly	2	Key symptoms	3	NS	0 .67	1176 (50.0%)	16.7 (1.0)	19.3%
Çifçili et al. (2010)	Random school sample	2	99.0%	3	DIS and DMS	1	Key symptoms	3	NS	0 .60	302 (76.5%)	17.3 (1.8)	20.2%
Dohnt et al. (2012)[4]	Stratified school sample	2	84.0%	3	DSM-IV and ICSD-III criteria	3	Multiple methods	3	NS	0 .73	384 (59.1%)	15.6 (1.0) *13-18*	7.8% DSM-IV (10.9% ICSD-III)
Fernandez-Mendoza et al. (2014)	Stratified sample	2	70.0%	3	DIS/ DMS, often/ very often	2	PBS	3	NS	0 .67	327 (46.2%)	9.2 (1.8) *5-12*	19.5%
Gehrman et al. (2011)[5]	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA	3	NS	0 .47	2824 (46.3%)	12.0 (2.6) *8-16*	Child = 19.5%, Parent = 6.6%

[1] "Poor sleep" rather than insomnia
[2] World Health Organisation, 2001
[3] Control group only
[4] DSM-IV criteria included in pooled prevalence estimates
[5] Parent and child ratings

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Goodwin et al. (2003)	School sample	2	30.6%	0	DIS/ DMS/ EMA/ NRS, frequently/ almost always	2	Sleep problems screening questionnaire	3	NS	0 .47	239 (55.2%)	*6-11*	25.5%
Huang et al. (2012)	Part of wider survey	3	NS	0	DIS/ DMS/ EMA during past 30 days	2	Key symptoms	3	k= .26[1]	1 .60	33692 (44.9%)	*13-15*	31.6%
Huang et al. (2010)	Random school sample	2	95.1%	3	DIS/ DMS/ EMA	1	PSQ	3	r= .64	2 .73	1906 (37.7%)	*12-18*	18.7%
Joo et al. (2005)	Random school sample	2	80.9%	3	DIS/ DMS/ EMA, 3+ times weekly, over past 4 weeks	3	Key symptoms	3	r= .73-.92	3 .93	3871 (69.8%)	16.8 (0.8)	12.7%
Kaneita et al. (2006)	Cluster sample of schools	2	64.8%	2	DIS/ DMS/ EMA, often/ always, over past month	3	Key symptoms	3	NS	0 .67	102451 (54.7%)	*12-18*	23.5%
Kilincaslan et al. (2014)	Cluster sample of schools	2	92.2%	3	SOL[2] >30mins/ DMS, several nights/ every night	2	Key symptoms	3	NS	0 .67	3485 (49.0%)	16.1 (1.0) *14-17*	24.1%

[1] k= test-retest reliability/ inter-rater reliability
[2] SOL= Sleep onset latency

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Kirmil-Gray et al. (1984)[1]	Health classes of three schools	2	NS	0	SOL 45+ mins/ 3+ night-wakings/ night-wakings 30+ mins, 3+ weekly	2	Key symptoms	3	NS	0 .47	277 (46.9%)	15.0 *13-17*	11.0%
Lee et al. (2012)	School sample	2	84.1%	3	ICD-10[2]	3	ICD-10	3	NS	0 .73	7172 (37.8%)	16.8 (1.1) *12-17*	9.7%
Liu et al. (2000)	Random/ stratified sample	2	97.0%	3	DIS/ DMS/ EMA, often, past month,	3	Key symptoms	3	NS	0 .73	1365 (60.3%)	14.6 (3.4) *12-18*	16.9%
Liu et al. (2008)[3]	Representative/ random school sample	2	97.8%	3	DIS/ DMS/ EMA, 3+ weekly, past month	3	Key symptoms	3	α= .5 k= .4	2 .87	1066 (58.6%)	Median= 14.5 *12-17*	16.1%
Mak et al. (2010)	Population survey of obesity	3	84.5%	3	DIS/ DMS/ EMA, past 30 days	2	Key symptoms	3	NS	0 .73	28839 (49.7%)	*12-18*	35.0%
Mak et al. (2012)	Population survey of obesity	3	94.8%	3	DIS/ DMS/ EMA, past 30 days	2	Key symptoms	3	k= .26	2 .87	22678 (41.6%)	*12-18*	21.5%
Morioka et al. (2013)	Random school sample. Part of wider survey	3	92.2%	3	DIS/ DMS/ EMA, always/ often, past 30 days,	3	Key symptoms	3	NS	0 .80	98867 (49.4%)	*12-18*	21.5%

[1] "Chronic poor sleepers"
[2] ICD-10= International Classification of Diseases (10th edition)
[3] 7th and 10th grade

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Munezawa et al. (2011)	Stratified school sample	2	87.2%	3	DIS/ DMS/ EMA, often/ always, over past month	3	Key symptoms	3	NS	0 .73	94777 (50.7%)	*12-18*	21.8%
Murray et al. (2012)[1]	Advertisements	2	56.0%	1	DIS/ DMS, often/ frequently/ always	2	Adolescent sleep-wake scale[2]	3	α= .81	3 .73	60 (33%)	15.1 (1.8) *12-18*	22.0%
Pan et al. (2012)	Random school sample	2	China= 95.1% Macau= 95.7%	3	ICSD-2	3	ICSD-2	3	NS	0 .73	China= 861 (47.6%) Macau= 618 (41.9%)	China = 15.3 (1.8) Macau = 15.6 (1.8)	China= 22.9% Macau= 16.5%
Patten et al. (2000)[3]	National survey of smoking	3	65.8%	2	DIS/ DMS, often, over 12 months	3	Key symptoms	3	NS	0 .73	7960 (50.7%)	*12-18*	14.4%
Radecki & Brunton (1993)[4]	National survey of office-based medical practices.	2	NS	0	Sleeplessness/ can't sleep/ trouble falling to sleep	1	Physician consultation	1	NS	0 .27	18325 (NS)	*<18*	0.46%
Roane et al. (2008)	Public use dataset	3	NS	0	DIS/ DMS, almost/ every day	2	Key symptoms	3	NS	0 .53	4494 (47.6%)	15.8 (1.5) *12-18*	9.4%

[1] Control group only
[2] LeBourgeois et al. (2005)
[3] Baseline only. Authors use the term “sleep problems” but employ the symptoms of insomnia to define this.
[4] Under 18 years only

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Roberts & Duong (2013)[1]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interviews based on DSM-IV	3	NS	0 .73	4175 (51.0%)	*11-17*	26.8%
Roberts et al. (2004)	Stratified school sample	2	67.0%	2	DMS/ DIS/ EMA/ NRS, often/ every day, over 4 weeks	3	DSM-IV criteria	3	NS	0 .67	5118 (NS)	Median= 15.0 *13-18*	12.3%
Roberts et al. (2001)	School sample	2	85.3%	3	DSM-IV	3	DSM Scale for Depression[2]	3	α>.9	3 .93	5423 (50.1%)	*10-17*	16.3%
Sharma et al. (2009)[3]	Stratified sample based on groundwater fluoride	1	NS	0	Lack of sleep	1	Measure distributed by doctor	1	NS	0 .20	999 (50.5%)	*6-18*	1.6%
Siomos et al. (2009)	Cross-section of schools	2	99.8%	3	ICD-10	3	AIS[4]	3	α= .82	3 .93	2195 (49.1%)	15.3 (1.7) *13-18*	11.4%
Steinsbekk & Wichstrøm (2015)	Weighted sample	1	79.5%	3	DSM-IV	3	DSM-IV	3	k= 0.9	3 .87	995 (49.1%)	4.4 (0.2) *4*	16.6%
Yen et al. (2008)	Stratified/ random school sample	2	72.0%	3	ICD-10	3	AIS	3	α= .67 r= .72	3 .93	8004 (47.8%)	14.7 (1.7) *12-18*	11.8%

[1] Baseline only
[2] Roberts, Roberts & Chen (1997)
[3] 6-18 years only
[4] AIS= Athens Insomnia Scale (Soldatos et al., 2000)

Table 1.6 - Summary of articles reporting the prevalence of insomnia in non-clinical samples

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Zhang et al. (2009)	Multiple schools	2	70.3%	3	DSM-IV/ ICSD-10	3	Diagnostic criteria	3	NS	0 .73	5695 (50.3%)	9.2 (1.8) *6-13*	4.0%
Zhang et al. (2011)[1]	Epidemiological study	2	47.2%	0	DIS/ DMS/ EMA/ 3+ weekly, over 12 months	3	Key symptoms	3	α= .87	3 .73	6447 (50.6%)	9.2 (1.8)	4.0%
Zhang et al. (2015)	Random school sample	2	76.4%	3	DIS/ DMS/ EMA/ 3+ weekly, over 12 months	3	Key symptoms	3	NS	0 .73	3086 (52.1%)	9.7 (1.7)/ 9.0 (1.6) *7-14*	3.7%

[1] Baseline data only

When comparing the articles described in Table 1.6, it was noted that the literature differed greatly across all elements of the quality framework, and the weightings and reported prevalence estimates varied widely as a result. Prevalence estimates ranged from 0.46% (Radecki & Brunton, 1993) to 35.0% (Mak et al., 2010), which suggests great variability in the methodology employed and the definition of the construct of insomnia, especially when considering that all of the samples were from non-clinical populations of a similar age-group.

Across the articles, the majority of the data was collected through questionnaire distribution at multiple schools. Although this posed a considerable risk for response bias, in terms of an increased interest from families for whom sleep was a concern, participant response rates were typically very high, with roughly half of the articles receiving the top weighting associated with a participant response rate greater than 70%. Other methods of sample recruitment that received high ratings included those that were conducting a wider survey on a construct other than sleep disorders, but had included items assessing for the presence of insomnia. This limited the likelihood of response bias. For example, the two articles by Mak and colleagues collected data on insomnia as part of a national survey of obesity. The authors included measures of smoking, alcohol consumption, exercise and health problems in addition to questions regarding sleep disorders, such as insomnia (Mak et al., 2010; Mak, Lee, Ho, Lo & Lam, 2012). This therefore reduced the likelihood of individuals with sleep problems being any more likely to return the questionnaire than other subgroups within the population. Other authors who employed similar methods of recruitment included Roberts and colleagues, who made use of the Teen Health 2000 dataset. This incorporated data on several

aspects of adolescent health and wellbeing, including sleep (Roberts & Duong, 2013; Roberts et al., 2008a; 2008b). However, only the 2013 article was retained for the pooled prevalence estimates, because it was concluded that the same sample had been used across the three publications.

Publications that had employed the use of wider datasets, designed to study an alternative construct of interest, sometimes only incorporated a single item to assess for insomnia, as part of a broader health questionnaire (such as Radecki & Brunton, 1993). This was in contrast to authors who had made use of standardised or well-validated measures specifically developed to explore the construct of sleep disorders, or even insomnia explicitly (such as Alvaro, Roberts & Harris, 2014). However, when comparing the content of the individual items themselves, they were often the same. As a result, articles which did not use a well-known measure specifically developed to assess for insomnia were able to receive the same rating as standardised measures, if it was evident that the items included in the chosen measure were a valid assessment of the construct of insomnia. Furthermore, articles where a single symptom had been used as the determinant of insomnia are included in the pooled meta-analyses for overall insomnia as well as the individual symptom.

The criteria employed by the articles to define when a participant met the threshold for insomnia differed between authors. Although the majority of papers considered presence of any of the three main insomnia symptoms as being indicative of insomnia, others were more stringent and required two or more symptoms to be present (Archbold, Pituch, Panahi & Chervin, 2002; Çifçili et al., 2010). Similarly, whilst several authors required the signs of insomnia to be

described as occurring frequently, often, or a minimum of three times a week, others simply required them to be present for participants to be classified as having current insomnia (Camhi, Morgan, Pernisco & Quan, 2000; Radecki & Brunton, 1993; Sharma, Sohu & Jain, 2009). Furthermore, the duration for which the symptoms needed to have persisted varied from 4 weeks to 12 months (Chen, Truong & Tsai, 2013; Huang, Ho, Lo, Lai & Lam, 2012; Joo et al., 2005; Patten, Choi, Gillin & Pierce, 2000; Zhang et al., 2011, 2015). These variations in the definition of insomnia may have accounted for some of the variability observed in the reported prevalence estimates. Prevalence estimates for the 44 papers described above were calculated based on the random- and the quality-effects (Figures 1.2 and 1.3).

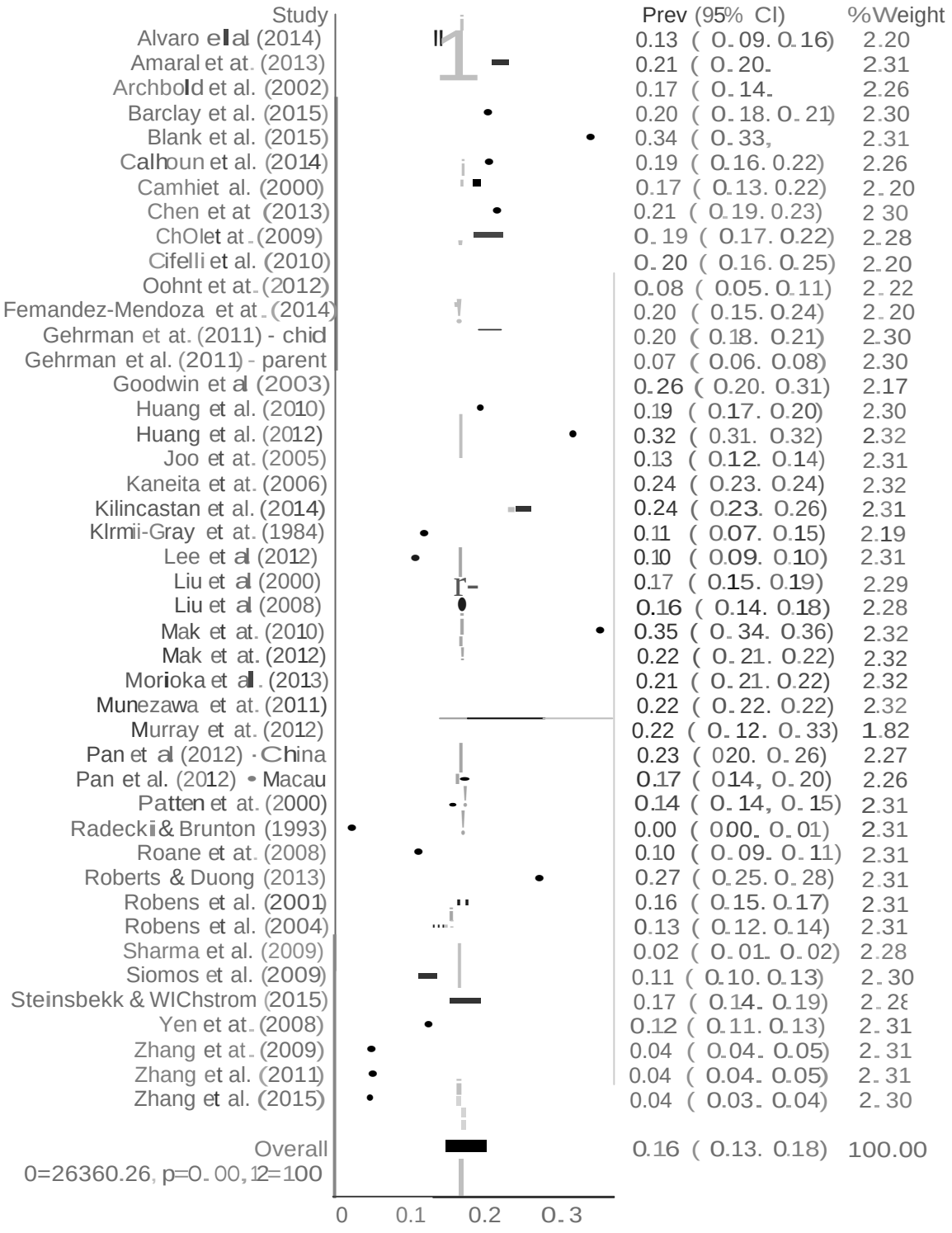

Figure 1.2 • Pooled estimates for the prevalence of insomnia. using a random-effects model.

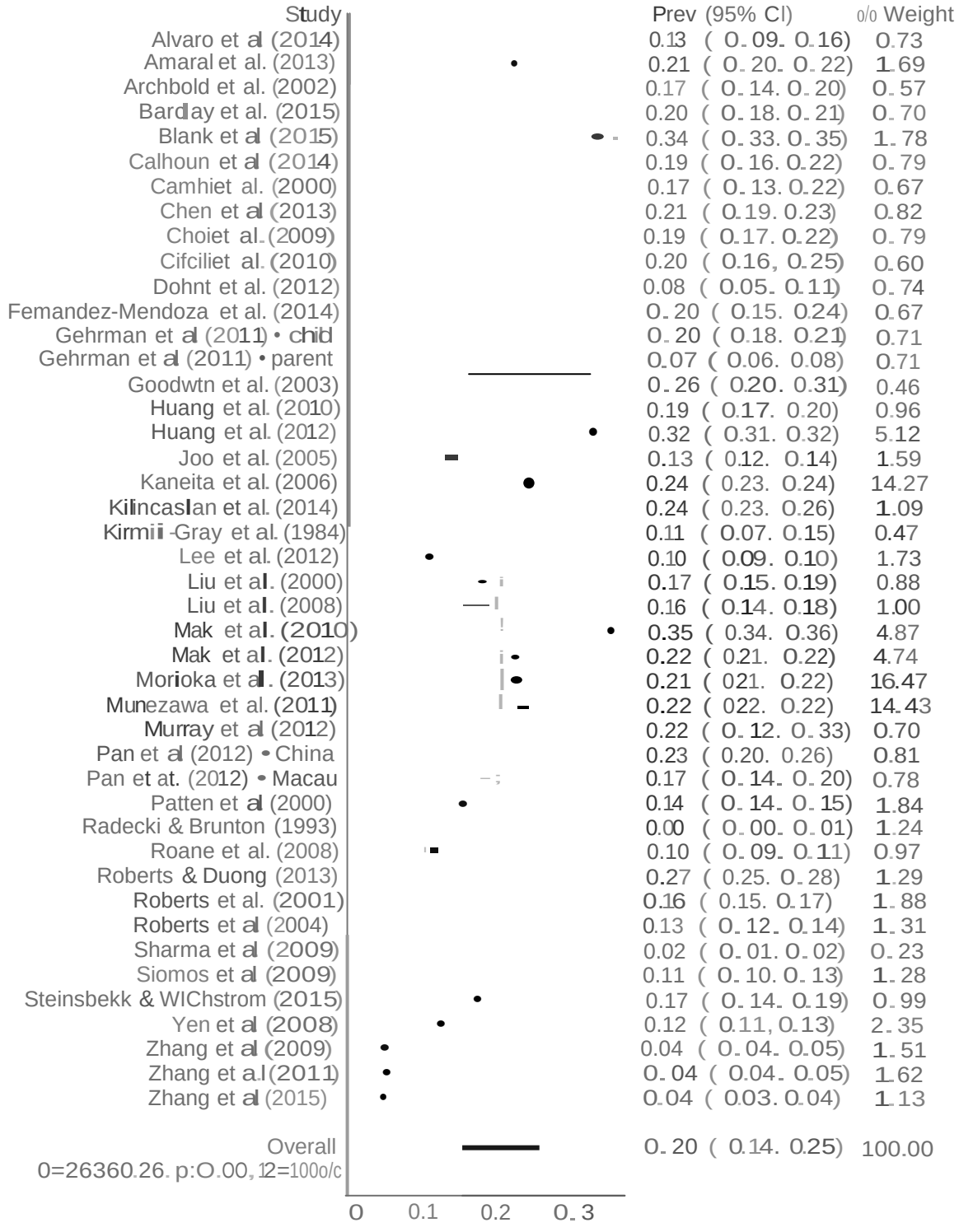

Figure 1.3. Pooled estimates for the prevalence of insomnia. using a quality-effects model.

The results of the pooled estimates suggested that the prevalence of insomnia is 15.7%[1] (CI= 13.1-18.5, I^2= 99.8) for the random-effects model, and 19.6% (CI= 14.5-25.3, I^2= 99.8) for the quality-effects model. The difference between these two estimates is fairly large, and it is interesting that the random-effects model has generated a more conservative prevalence statistic. This suggests that some of the studies receiving lower scores in the quality weightings reported lower prevalence rates than those receiving higher quality weightings. These studies receiving lower quality ratings have therefore biased the random-effects prevalence estimate. It should be noted that the I^2 statistic is 100% ($p < .1$) which indicates a very high level of heterogeneity within the meta-analysis, to the extent that it is unlikely to have occurred by chance. This may be due to methodological differences, or difference in the samples employed by the different studies.

It was considered that one potential cause of variance in the prevalence estimates may be due to differences in sleep quality over childhood and adolescence. As a result, articles that constituted a sample of adolescents, aged 12-18 years, were analysed separately to articles employing participants aged 4-12 years, to see if the heterogeneity observed in the overall pooled prevalence estimates would be reduced. It was found that the prevalence of insomnia in adolescents according to the random-effects model was 18.1 % (CI= 16.0-20.3, I^2= 99.7), and 21.3% (CI= 17.7-25.1, I^2= 99.7) according to the quality-effects model. The pooled prevalence data for the participants under the age of 12 years was 19.7% (CI= 16.5-23.0, I^2= 70.1) in the random-effects model, and 18.6% (CI= 15.3-

[1] Please note that the estimates reported in the forest plots are given as decimals, whereas the estimates given in the text are given as percentages correct to one decimal place.

22.1, I^2= 70.1) in the quality-effects model. This suggests that there is little effect of age in the variance of the data, although there is slightly less heterogeneity in the data for the younger age group.

Insomnia with evidence of daytime impairment. During the application of the quality framework to the articles, it was noticed that a number of articles included an additional criterion into their definitions of insomnia; that it must occur alongside impairment in daytime functioning, or cause some level of distress. Despite many articles citing one of the editions of the DSM or ICSD publications in the development of their definition, and this criterion being stated in said publications, authors did not then assess for this in their chosen measures. Articles in which it was explicitly stated that this element was included in either the definition or assessment of insomnia are summarised in the table below (Table 1.7). For some of these articles, the prevalence estimate of insomnia including daytime impairment is equivalent to that given in Table 1.6, but for others, this prevalence statistic was reported separately, and so differs from any reported previously.

Twelve articles were identified that reported a prevalence figure for insomnia with daytime impairment. Three were then excluded as they employed duplicate samples (Amaral, de Figueiredo Pereira, Silva Martins, de Serpa & Sakellarides, 2013; Roberts et al., 2006, 2008b). The remaining nine articles are summarised in Table 1.7. Reported prevalence estimates ranged from 6.7-19.5% (Barclay, Gehrman, Gregory, Eaves & Silberg., 2015; Roberts & Duong, 2013).

Table 1.7 - Summary of articles reporting the prevalence of insomnia with daytime impairment in non-clinical samples

Authors	Quality ratings											N % Male	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Alvaro et al. (2014)	Multiple schools	2	NS	0	DSM-IV[1]	3	ISI[2]	3	α= .83 r= .79[3]	3	.73	318 (51.6%)	15.0 (1.3) *12-18*	12.5%
Amaral et al. (2014)	Multiple schools	2	82.1%	3	EMA[4], 3+ weekly, past month	3	DSM-IV	3	NS	0	.73	6919 (47.0%)	*12-18*	8.3%
Barclay et al. (2015)[5]	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA[6]	3	NS	0	.47	2789 (46.0%)	Mode= 8.3 *8-17*	19.5%
Chung et al. (2014)	Stratified school sample	2	72.0%	3	DSM-IV	3	Insomnia Interview Schedule[7]	3	NS	0	.73	290 (43.8%)	14.4 (1.4) *12-18*	9.3%
Johnson et al. (2006)	Health insurance database	3	71.2%	3	DSM-IV	3	DSM-IV/ ICSD-R[8]	3	NS	0	.80	1014 (49.7%)	*13-16*	9.4%
Roberts & Duong (2013)[9]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interviews based on DSM-IV	3	NS	0	.73	4175 (51.0%)	*11-17*	6.7%

[1] DSM= Diagnostic and Statistical Manual
[2] ISI= Insomnia Severity Index (Morin,1993)
[3] α= Cronbach's alpha; r= test-retest reliability/ inter-rater reliability
[4] EMA= Early morning awakening
[5] Wave 1 only
[6] CAPA= Child and Adolescent Psychiatric Assessment (Angold et al., 1995)
[7] Morin (1993)
[8] ICSD-R= International Classification of Sleep Disorders (revised)
[9] Baseline only.

Table 1.7 - Summary of articles reporting the prevalence of insomnia with daytime impairment in non-clinical samples

Authors	Quality ratings											N % Male	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Roberts et al. (2008)[1]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interviews based on DSM-IV	3	NS	0	.73	4175 (51.1%)	*11-17*	7.2%
Siomos et al. (2009)	Cross-section of schools	2	99.8%	3	ICD-10[2]	3	AIS[3]	3	α= .82	3	.93	2195 (49.1%)	15.3 (1.7) *13-18*	11.4%
Yen et al. (2008)	Stratified/ random school sample	2	72.0%	3	ICD-10	3	AIS	3	α= .67 r= .72	3	.93	8004 (47.8%)	14.7 (1.7) *12-18*	11.8%

[1] Wave 1 only.
[2] ICD-10= International Classification of Diseases (10th edition)
[3] AIS= Athens Insomnia Scale (Soldatos et al., 2000)

Prevalence data and the quality weighting for each article were then entered into the pooled meta-analyses. The random- and quality-effects models are summarised in Figure 1.4.

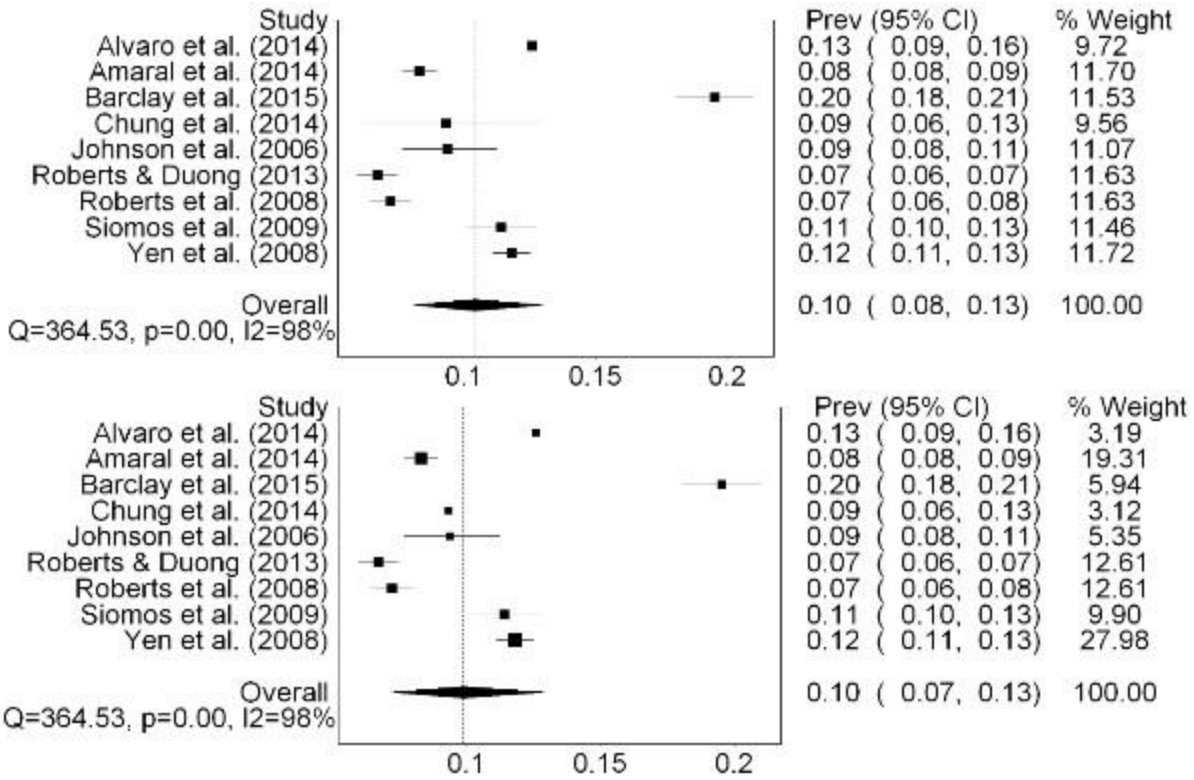

Figure 1.4 - Pooled estimates for the prevalence of insomnia with daytime impairment, using random-effects (upper panel) and quality-effects (lower panel) models.

The pooled prevalence estimates for insomnia with daytime impairment were 10.4% (CI= 8.0-13.0) and 9.9% (CI= 7.1-13.0) for the random- and quality-effects models respectively. As expected, the added criterion of the presence of daytime impairment, in addition to the symptoms of insomnia, demonstrated a decrease in the generated pooled prevalence estimates. As discussed previously, the presence of some form of negative daytime consequence to the experience of insomnia is denoted in the criteria set out by both the most recent DSM and ICSD publications.

Clinically, this would seem important, as the absence of such consequences implies that any observed signs of sleeplessness may be artefacts of normal variation in a child's sleeping pattern, and therefore not indicative of a clinical problem. Yet, this element of the diagnostic criteria is less commonly mentioned in the assessment phase of research studies. For example, the frequently-used Athens Insomnia Scale (AIS; Soldatos, Dikeos & Paparrigopoulos, 2000) includes items on each of the symptoms of insomnia, as well as items exploring daytime impairment in wellbeing and functioning. However, it is possible to score above the recommended cut-off, without scoring on the latter items, and therefore it is unclear whether researchers are only considering participants scoring positively on these items as experiencing insomnia, or whether they are using the total score across any item to define the disorder.

Insomnia with the exclusion of additional comorbidities. In addition to the added criterion of daytime impairment, some articles required insomnia to occur independently of other comorbidities, such as another sleep disorder. In the ICSD, this would be referred to as idiopathic insomnia, differentiated from psychophysiological insomnia or extrinsic sleep disorders, such as poor sleep hygiene or sleep-onset association. In the DSM, this would be referred to as primary insomnia, which differs from insomnia related to a mental or physical health disorder, or substance use. Two articles were excluded due to non-unique samples (Roberts, et al., 2006, 2008b), leaving four papers remaining (Table 1.8).

Table 1.8 - Summary of articles reporting the prevalence of insomnia with exclusion of comorbidities in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Ohayon & Roberts (2001)[1]	Population/ stratified sample	2	78.0%	3	DSM-IV/ ICSD[2]	3	Sleep-EVAL	3	k= .78-.71[3]	3	.93	794 (54.1%)	*15-18*	3.3%
Ohayon et al. (2000)[4]	Stratified sample	2	80.8%	3	DSM-IV/ ICSD	3	Sleep-EVAL	3	k= .70-.78	3	.93	1125 (54.1%)	*15-18*	2.2%
Roberts & Duong (2013)[5]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interview based on DSM-IV	3	NS	0	.73	4175 (51.0%)	*11-17*	4.7%
Robert et al. (2008)[6]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interview based on DSM-IV	3	NS	0	.73	4175 (51.1%)	*11-17*	4.7%

[1] 15-18 years only
[2] DSM= Diagnostic and Statistical Manual; ICSD= International Classification of Sleep Disorders
[3] k= test-retest reliability/ inter-rater reliability
[4] 15-18 years only
[5] Baseline only
[6] Wave 1 only

Prevalence data and the quality weighting for each article were entered into the pooled meta-analyses. The random- and quality-effects models are summarised in Figure 1.5.

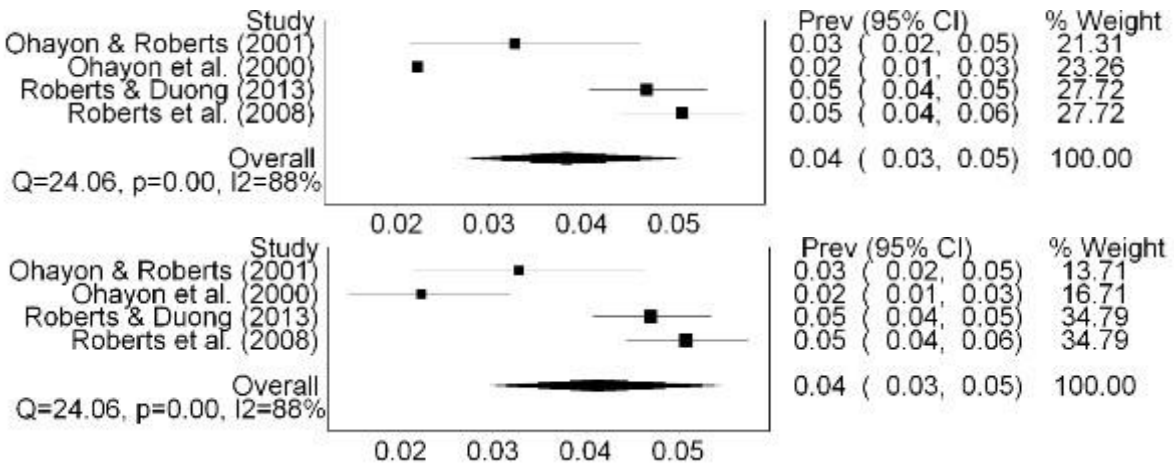

Figure 1.5 - Pooled estimates for the prevalence of insomnia with the exclusion of comorbidities, using random-effects (upper panel) and quality-effects (lower panel) models.

The pooled prevalence estimates for insomnia without comorbidities were 3.8% (CI= 2.7-5.1) and 4.1% (CI= 3.0-5.5) for the random- and quality-effects models respectively. It can be seen that, in comparison to the prevalence rates of insomnia in Figure 1.4, excluding cases where symptoms of insomnia had arisen due to additional factors, prevalence rates decreased by approximately 80%. This implies that the vast majority of cases of insomnia are not idiopathic, but are related to a comorbid condition. This is interesting when considering that the DSM-IV does not allow for a diagnosis of insomnia in the presence of an additional disorder, but would instead suggest that any evidence of insomnia is secondary to the comorbidity. The exception to this would be if the insomnia appeared to be causal, or at least strongly contributing to the additional disorder. In terms of the existing

research literature, the majority of authors base their definitions of insomnia on DSM-IV criteria, yet do not attempt to control for additional comorbidities. Therefore, whilst the symptoms of insomnia reported by their participants may be clinically important, they do not necessarily meet the recommended thresholds. However, it is important for clinicians to be aware of the types of disorders in which they are more likely to observe insomnia, whether it primary or secondary to the comorbidity.

Difficulty initiating sleep. Forty-nine articles were identified that reported a prevalence figure for difficulty initiating sleep (DIS), also referred to as onset insomnia or early insomnia, after three had been removed for duplicated samples (Liu, 2004; Liu & Zhou, 2002; Robert, Roberts & Chan, 2006) and one for failing to score on the three key criteria (Lazaratou, Dikeos, Anagnostopoulos, Sboukou & Soldatos, 2005). These are summarised in Table 1.9. Reported prevalence estimates for the remaining articles ranged from 2.1-62.0% (Nevéus, Läckgren, Stenberg, Tuvemo & Hetta, 1998; Siomos et al., 2009).

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Amaral et al. (2013)	Multiple schools	2	82.1%	3	DSM-IV[1]	3	DSM-IV	3	NS	0 .73	6919 (47.0%)	*12-18*	8.9%
Barclay et al. (2015)[2]	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA[3]	3	NS	0 .47	2789 (46.0%)	Mode= 8.3 *8-17*	14.6%
Blader et al. (1997)	Random school sample	2	60.0%	2	DIS[4], 3+ weekly	2	Key symptom	3	NS	0 .60	972 (NS)	7.5 (1.1) *5-12*	11.3%
Blank et al. (2015)	National survey	3	82.9%	3	DSM-IV	2	Interviews based on DSM-IV	3	NS	0 .73	6483 (48.6%)	*13-18*	24.5%
Calhoun et al. (2014)	Population/ stratified school sample	2	70.0%	3	ICSD[5]	3	PBS[6]	3	NS	0 .73	700 (46.0%)	8.8 (1.8) *5-12*	14.3%
Chen et al. (2013)[7]	National survey, stratified sample	3	NS	0	DIS, often/ always, past 4 weeks	3	Insomnia self-assess. inventory[8]	3	NS	0 .60	2113 (52.9%)	*15-17*	3.2%
Chung & Cheung (2008)[9]	Multiple schools	2	86.0%	3	DIS, 3+ weekly, past 3 months	3	Sleep Quality Index[10]	3	r= .55[11]	2 .87	1335 (NS)	*12-16*	5.4%

[1] DSM= Diagnostic and Statistical Manual
[2] Wave 1 only
[3] CAPA= Child and Adolescent Psychiatric Assessment (Angold et al., 1995)
[4] DIS= Difficulty initiating sleep
[5] ICSD= International Classification of Sleep Disorders
[6] PBS= Pediatric Behavior Scale (Lindgren & Koeppl, 1987)
[7] "Poor sleep" rather than insomnia
[8] World Health Organisation, 2001
[9] 12-16 years only
[10] Urponen et al., 1991
[11] r= test-retest reliability/ inter-rater reliability

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings											N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Çifçili et al. (2010)	Random school sample	2	99.0%	3	DIS	1	Key symptoms	3	NS	0	.60	302 (76.5%)	17.3 (1.8)	32.8%
Choi et al. (2009)[1]	Random/ stratified school sample	2	86.7%	3	DIS 3+ weekly	2	Key symptoms	3	NS	0	.67	1176 (50.0%)	16.7 (1.0)	12.0%
Chung et al. (2014)	Stratified school sample	2	72.0%	3	DSM-IV[2]	3	Insomnia Interview Schedule[3]	3	NS	0	.73	290 (43.8%)	14.4 (1.4) *12-18*	35.9%
Gehrman et al. (2011)	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA	3	NS	0	.47	2824 (46.3%)	12.0 (2.6) *8-16*	Child = 14.5%, Parent = 4.9%
Huang et al. (2012)	Stratified school sample. Part of wider survey	3	NS	0	DIS, past 30 days	2	Key symptoms	3	k= .26[4]	1	.60	33692 (44.9%)	*13-15*	17.2%
Huang et al. (2010)	Random school sample	2	95.1%	3	DIS	1	PSQ[5]	3	r= .64	2	.73	1906 (37.7%)	*12-18*	24.7%
Johnson et al. (2004)[6]	Existing dataset from random sample	3	NS	0	Does not fall asleep easily	1	DISC-1[7]	3	r= .24	1	.53	749 (NS)	14.0 (3.0)	34.3%

[1] Control group only
[2] DSM= Diagnostic and Statistical Manual (4th edition; 3rd edition – revised)
[3] Morin (1993)
[4] k= test-retest reliability/ inter-rater reliability
[5] PSQ= Pediatric Sleep Questionnaire (Chervin et al., 2000)
[6] 14 years only
[7] DISC= Diagnostic Interview Schedule for Children

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Johnson et al. (2006)	Health insurance database	3	71.2%	3	DSM-IV	3	Interviews based on DSM-IV and ICSD-R	3	NS	0 .80	1014 (49.7%)	*13-16*	11.6%
Junker et al. (2014)[1]	Population sample for non-sleep study	3	88.0%	3	DIS, often/ almost every night, past month	3	Key symptoms	3	NS	0 .73	4325 (N.s)	13-16	8.0%
Kaneita et al. (2006)	Cluster sample of schools	2	64.8%	2	DIS, often/ always, past month	3	Key symptoms	3	NS	0 .67	102451 (54.7%)	*12-18*	14.8%
Kilincaslan et al. (2014)	Cluster sample of schools	2	92.2%	3	SOL[2] > 30mins	2	Key symptoms	3	NS	0 .67	3485 (49.0%)	16.1 (1.0) *14-17*	12.4%
Kirmil-Gray et al. (1984)	Health classes of three schools	2	NS	0	DIS, 3+ weekly	2	Key symptoms	3	NS	0 .47	277 (46.9%)	15.0 *13-17*	13.8%
Lee et al. (2012)	School sample	2	84.1%	3	ICD-10[3]	3	ICD-10	3	NS	0 .73	7172 (37.8%)	16.8 (1.1) *12-17*	6.5%
Leger et al. (2012)	National survey	3	93.5%	3	DIS, every night/ several times weekly, past 6 months	3	Key symptom	3	NS	0 .80	9251 (50.2%)	*11-15*	19.2%

[1] Under 16 years only
[2] SOL= sleep onset latency
[3] ICD-10= International Classification of Diseases (10th edition)

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Lehmkuhl et al. (2008)	Random school sample of new-starters	2	29.0%	0	DIS, often	2	Key symptom	3	NS	0 .47	1388 (51.3%)	5.5	5.1%
Liu et al. (2000)	Representative/ random sample	2	97.0%	3	DIS, often, past month	3	Key symptoms	3	NS	0 .73	1365 (60.3%)	14.6 (3.4) *12-18*	10.8%
Liu et al. (2008)[1]	Representative/ random sample	2	97.8%	3	DIS, 3+ weekly, past month	3	Key symptoms	3	α= .5[2] k= .4	2 .87	1066 (58.6%)	Median= 14.5 *12-17*	7.4%
Mak et al. (2010)	Population survey of obesity	3	84.5%	3	DIS, past 30 days	2	Key symptoms	3	NS	0 .73	28839 (49.7%)	*12-18*	19.0%
Mak et al. (2012)	Population survey of obesity	3	94.8%	3	DIS, past 30 days	2	Key symptoms	3	k= .26	2 .87	22678 (41.6%)	*12-18*	12.3%
Morioka et al. (2013)	Random school sample. Part of wider survey	3	92.2%	3	DIS, always/ often, past 30 days	3	Key symptoms	3	NS	0 .80	98867 (49.4%)	*12-18*	13.3%
Munezawa et al. (2009)	Multiple schools	2	78.7%	3	SOL > 30 mins, weekly+, past month	3	PSQI[3]	3	NS	0 .73	916 (62.0%)	*12*	6.8%
Murray et al. (2012)[4]	Advertisements	2	56.0%	1	DIS often/ frequently/ always	2	Adolescent sleep-wake scale[5]	3	α= .81	3 .73	60 (33%)	15.1 (1.8) *12-18*	18.6%

[1] 7th and 10th grades only
[2] α= Cronbach's
[3] PSQI= Pittsburgh Sleep Quality Index
[4] Control group only
[5] LeBourgeois et al. (2005)

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings											N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Nevéus et al. (2001)	Representative school sample	2	74.0%	3	DIS, nightly/ weekly	2	Key symptom	3	NS	0	.67	1413 (49.3%)	7.9 *6-11*	13.9%
Nevéus et al. (1999)[1]	Multiple schools	2	74.0%	3	"Onset insomnia", nightly/ weekly/ monthly	2	Measure designed to study sleep problems	2	NS	0	.60	1293 (53.0%)	7.9 *6-10*	51.0%
Nevéus et al. (1998)[1]	Single school	1	91.0%	3	"Onset insomnia", nightly/ weekly/ monthly	2	Measure designed to study sleep problems	2	NS	0	.53	100 (49.0%)	8.7 (1.0) *7-10*	62.0%
Ohayon & Roberts (2001)[2]	Population/ stratified sample	2	78.0%	3	DSM-IV or ICSD	3	Sleep-EVAL	3	k= .71-.78	3	.93	794 (54.1%)	*15-18y*	14.1%
Ohida et al. (2004)	Stratified school sample. Part of wider data collection.	3	87.0%	3	DIS, always/ often	2	Key symptom	3	NS	0	.73	106295 (51.3%)	*12-18*	15.6%
Pagel et al. (2007)	Two schools	2	47.6%	0	DIS, always/ every night	2	PSQ	3	NS	0	.47	165 (49.1%)	14.2 (1.5) *11-17*	21.2%
Pallesen et al. (2008)	National survey	3	58.0%	1	Sleep-onset difficulties, daily/ 1+ weekly, over 6 months	3	Key symptom	3	NS	0	.67	4711 (51.8%)	11-15	20.4%

[1] Control group only
[2] 15-18 years only

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Pan et al. (2012)	Random school sample	2	China= 95.1% Macau= 95.7%	3	ICSD-2	3	ICSD-2	3	NS	0 .73	China= 861 (47.6%) Macau= 618 (41.9%)	China = 15.3 (1.8) Macau = 15.6 (1.8)	China= 17.2% Macau= 10.2%
Roberts et al. (2004)	Stratified school sample	2	67.0%	2	DIS, often/ every day, past 4 weeks	3	DSM-IV	3	NS	0 .67	5118 (NS)	Median= 15.0 *13-18*	16.7%
Roberts et al. (2008)[1]	Epidemiological study	3	66.0%	2	DSM-IV	3	Intervie ı based on DSM-IV	3	NS	0 .73	4175 (51.1%)	*11-17*	7.1%
Roberts et al. (2002)	Epidemiological study	3	NS	0	DIS, almost every day, past 4 weeks	3	DS I-IV	3	α>.9	3 .80	4175 (50.8%)	*11-17*	6.5%
Shang et al. (2005)	Stratified school sample	2	91.6%	3	DIS, 3+ weekly, over past month	3	SHQ[2]	3	k= .54-.85	3 .90	1391 (52.4%)	7.4 (1.5) *4-9*	18.5%
Shur-Fen (2006)	Random school sample	2	95.3%	3	DIS, 30+ mins, 3+ weekly, past 6 months	3	SHQ	3	NS	0 .73	2463 (52.4%)	11.6 (2.6) *6-16*	4.6%
Singareddy et al. (2013)[3]	Population/ stratified school sample	2	70.0%	3	DIS, very often, past 2 months	3	Key symptoms	3	NS	0 .73	666 (48.0%)	8.8 (1.7) *5-12*	13.3%

[1] Wave 1 only
[2] SHQ= Sleep Habits Questionnaire (Gau, 2000)
[3] Non-self-harming group only

Table 1.9 - Summary of articles reporting the prevalence of difficulty initiating sleep in non-clinical samples.

Authors	Quality Ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Siomos et al. (2009)	Cross-section of schools	2	99.8%	3	ICD-10	3	AIS[1]	3	α .82	3 .93	2195 (49.1%)	15.3 (1.7) *13-18*	2.1%
Stein et al. (2001)	Paediatric clinics	1	NS	0	DIS >30 mins, 7x weekly	2	Key symptoms	3	NS	0 .40	472 (53.6%)	*4-12*	8.3%
Yang et al. (1987)	Multiple schools	2	NS	0	SOL >1hr, 4+ monthly, past 6 months	3	Key symptoms	3	NS	0 .53	846 (49.2%)	14.7 (1.4) *12-18*	14.9%
Zhang et al. (2009)	Multiple schools	2	70.3%	3	DSM-IV and ICSD-1	3	DSM-IV and ICSD-1	3	NS	0 .73	5695 (50.3%)	9.2 (1.8) *6-13*	2.5%
Zhang et al. (2011)[2]	Epidemiological study	2	47.2%	0	DIS, 3+ weekly, past 12 months	3	Key symptoms	3	α= .87	3 .73	6447 (50.6%)	9.2 (1.8)	2.5%
Zhang et al. (2015)	Random school sample	2	76.4%	3	DIS, 3+ weekly, past 12 months	3	Key symptoms	3	NS	0 .73	3086 (52.1%)	9.7 (1.7)/ 9.0 (1.6) *7-14*	2.9%

[1] AIS= Athens Insomnia Scale (Soldatos et al., 2000)
[2] Baseline data only

Many of the methodological observations made for the group of articles describing the prevalence of insomnia, are also relevant for the articles describing the prevalence of DIS. Sample recruitment primarily took place through schools, with efforts made to ensure a randomised, representative or stratified sample (Examples are Calhoun, Fernandez-Mendoza, Vgontzas, Liao & Bixler, 2014; Choi et al., 2009; Liu, Uchiyama, Okawa & Kurita, 2000; Singareddy et al., 2013). For example, Calhoun et al. (2014) distributed questionnaires through schools, and then selected a sample of 700 families, stratified by age, gender and risk of sleep-disordered breathing. Similarly, Choi and colleagues (2009) randomly selected schools stratified across nine geographical areas, to ensure the sample was representative of the population. Articles that received lower scores for sample recruitment included Barclay et al., (2015), and Gehrman et al. (2011), due to the samples consisting of twin pairs. Similarly, Stein, Mendelsohn, Obermeyer, Amromin and Benca (2001), and Nevéus et al. (1998) recruited from a restricted and non-random population, of a single school or paediatric clinic, therefore it is possible that the prevalence estimates gained from these samples may be less representative of the wider population.

The variety of definitions of DIS also reflect those discussed previously in the insomnia prevalence analysis, in terms of frequency and chronicity of symptoms. One difference, however, is that sleep onset latency (SOL) was also included here. Several articles suggested that a SOL of 30 minutes signified a difficulty in falling asleep (Kilincaslan, Yilmaz, Oflaz & Aydin, 2014; Munezawa, Kaneita, Yokoyama, Suzuki & Ohida, 2009; Shur-Fen, 2006; Stein et al., 2001), whereas Yang, Zuo and Eaton (1987) recommended that SOL was over an hour long. Other authors used

items questioning whether the child ‘falls asleep easily’ (Johnson, Cohen, Kasen, First & Brook, 2004). The presence of DIS was also more frequently assessed using a single item, whereas insomnia was often assessed over numerous items. As a result, one would expect the prevalence of insomnia to be greater than that of DIS, when considering that insomnia was frequently defined as the presence of any one symptom, one of which could include DIS. The reported prevalence estimates and quality ratings were entered into random-effects and quality-effects models to estimate the pooled prevalence statistics (Figures 1.6 and 1.7).

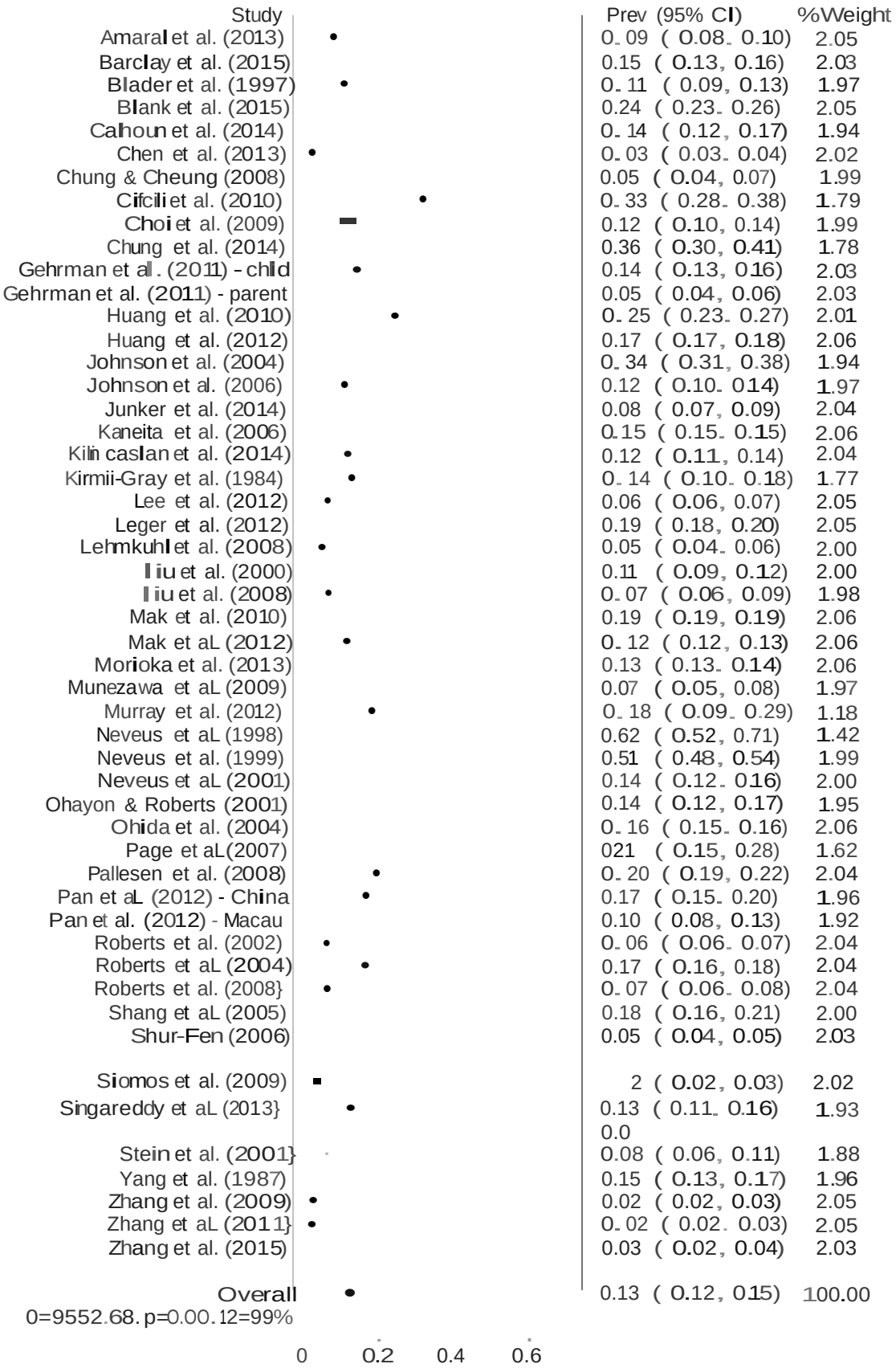

Figure 1.6- Pooled estimates for the prevalence of difficulty initiating sleep using a random-effects model.

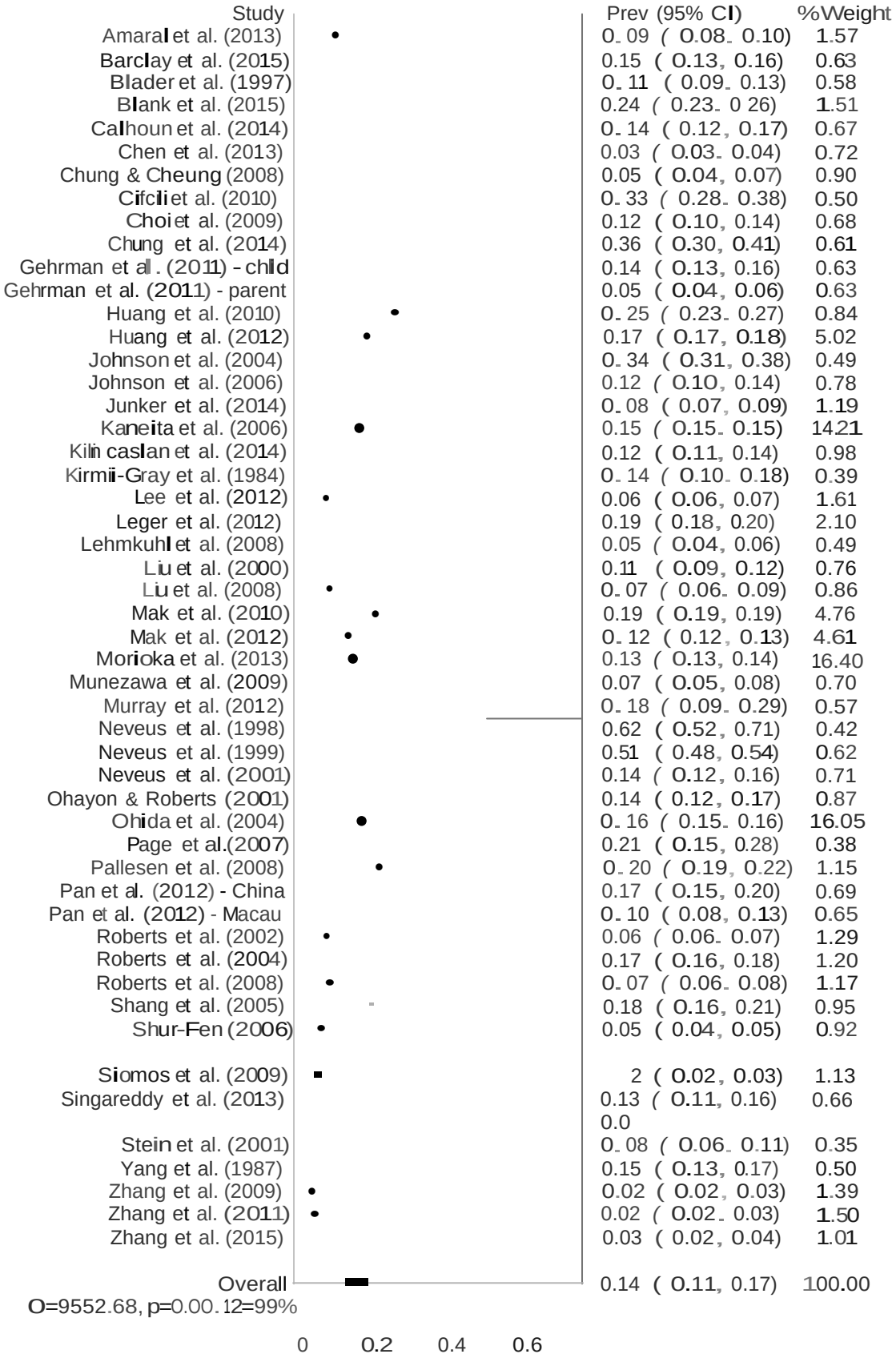

Figure 1.7- Pooled estimates for the prevalence of difficulty initiating sleep using a quality-effects model.

The results of the pooled prevalence analyses suggested that the prevalence of DIS is 13.2% (CI= 11.8-14.7) for the random-effects model, and 13.6% (CI= 10.9-16.7) for the quality-effects model. Visual inspection of the forest plot in both Figures 1.6 and 1.7 suggests that the articles by Nevéus et al. (1998, 1999) may be anomalous in comparison to the remainder of the articles. When looking in detail at these two articles, it can be seen that the authors defined DIS as being present when occurring at a minimum of once a month. This definition is considerably less conservative than the remaining articles. With these articles removed from the analysis, the prevalence estimate taken from the random-effects model was 12.1% (CI= 10.8-13.4), and 13.3% (CI= 10.7-16.2) in the quality-effects model. The considerable overlap between the confidence intervals for the original meta-analyses, and the meta-analyses with the Nevéus et al. articles removed suggests that there is no significant difference in the pooled prevalence estimates, and therefore the original estimates were retained.

Difficulty maintaining sleep. Forty-five articles were identified that reported a prevalence figure for difficulty maintaining sleep (DMS), also referred to in the literature as middle insomnia or night-waking, after three had been removed for non-unique samples (Liu, 2004; Liu & Zhou, 2002; Roberts et al., 2006), and one for failing to score on the three key criteria (Lazaratou et al., 2005). These are summarised in Table 1.10. Reported prevalence estimates for the remaining articles ranged from 1.3-64.2% (Johnson et al., 2004; Zhang et al., 2009, 2011, 2015).

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Amaral et al. (2013)	Multiple schools	2	82.1%	3	DSM-IV[1]	3	DSM-IV	3	NS	0 .73	6919 (47.0%)	*12-18*	6.1%
Barclay et al. (2015)[2]	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA[3]	3	NS	0 .47	2789 (46.0%)	Mode= 8.3 *8-17*	19.6%
Blader et al. (1997)	Random school sample	2	60.0%	2	DMS[4], 3+ weekly	2	Key symptom	3	NS	0 .60	972 (NS)	7.5 (1.1) *5-12*	6.5%
Blank et al. (2015)	National survey	3	82.9%	3	DSM-IV	3	Interviews based on DSM-IV	3	NS	0 .73	6483 (48.6%)	*13-18*	14.2%
Calhoun et al. (2014)	Population/ stratified school sample	2	70.0%	3	ICSD[5]	3	PBS[6], based on ICSD	3	NS	0 .73	700 (46.0%)	8.8 (1.8) *5-12*	10.7%
Chen et al. (2013)	National survey, stratified sample	3	NS	0	DMS, often/ always, past 4 weeks	3	Insomnia self-assess. inventory[7]	3	NS	0 .6	2113 (52.9%)	*15-17*	1.4%
Choi et al. (2009)[8]	Random/ stratified school sample	2	86.7%	3	DMS 3+ weekly	2	Key symptoms	3	NS	0 .67	1176 (50.0%)	16.7 (1.0)	7.6%

[1] DSM= Diagnostic and Statistical Manual
[2] Wave 1 only
[3] CAPA= Child and Adolescent Psychiatric Assessment (Angold et al., 1995)
[4] DMS= Difficulty maintaining sleep
[5] ICSD= International Classification of Sleep Disorders
[6] PBS= Pediatric Behavior Scale (Lindgren & Koeppl, 1987)
[7] World Health Organisation, 2001
[8] Control group only

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Chung & Cheung (2008)[1]	Multiple schools	2	86.0%	3	DMS 3+ weekly, past 3 months	3	Sleep Quality Index[2]	3	r= .52[3]	2 .87	1335 (NS)	*12-16*	6.8%
Chung et al. (2014)	Stratified schools sample	2	72.0%	3	DSM-IV	3	Insomnia Interview Schedule[4]	3	NS	0 .73	290 (43.8%)	14.4 (1.4) *12-18*	9.3%
Çifçili et al. (2010)	Random school sample	2	99.0%	3	DMS	1	Key symptoms	3	NS	0 .60	302 (76.5%)	17.3 (1.8)	45.0%
Gehrman et al. (2011)	Longitudinal twin study	1	NS	0	DSM-III-R[3]	3	CAPA	3	NS	0 .47	2824 (46.3%)	12.0 (2.6) *8-16*	Child = 19.6%, parent = 6.3%
Huang et al. (2012)	Stratified school sample. Part of wider survey	3	NS	0	DMS, past 30 days.	2	Key symptoms	3	k= .26[3]	1 .60	33692 (44.9%)	*13-15*	12.2%
Huang et al. (2010)	Random school sample	2	95.1%	3	DMS	1	PSQ[5]	3	r= .64	2 .73	1906 (37.7%)	*12-18*	7.4%

[1] 12-16 years only
[2] Urponen et al., 1991
[3] k or r= test-retest reliability/ inter-rater reliability
[4] Morin (1993)
[5] PSQ= Pediatric Sleep Questionnaire (Chervin et al., 2000)

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Johnson et al. (2004)[1]	Existing dataset from random sample	3	NS	0	Wakes in night/ wakes early in morning	1	DISC-1[2]	3	r= .24	1 .53	749 (NS)	14.0 (3.0)	64.2%
Johnson et al. (2006)	Health insurance database	3	71.2%	3	DSM-IV	3	Interviews based on DSM-IV and ICSD-R	3	NS	0 .80	1014 (49.7%)	*13-16*	4.5%
Kaneita et al. (2006)	Cluster sample of schools	2	64.8%	2	DMS, often/ always, past month	3	Key symptoms	3	NS	0 .67	102451 (54.7%)	*12-18*	11.3%
Kilincaslan et al. (2014)	Cluster sample of schools	2	92.2%	3	DMS, several nights/ every night	2	Key symptoms	3	NS	0 .67	3485 (49.0%)	16.1 (1.0) *14-17*	10.7%
Kirmil-Gray et al. (1984)	Health classes of three schools	2	NS	0	DMS, 3+ weekly	2	Key symptoms	3	NS	0 .47	277 (46.9%)	15.0 *13-17*	13.1%
Lee et al. (2012)	School sample	2	84.1%	3	ICD-10[3]	3	ICD-10	3	NS	0 .73	7172 (37.8%)	16.8 (1.1) *12-17*	4.5%
Lehmkuhl et al. (2008)	Random school sample of new-starters	2	29.0%	0	DMS, often	2	Key symptoms	3	NS	3 .47	1388 (51.3%)	5.5	5.2%

[1] 14 years only
[2] DISC-1= Diagnostic Interview Schedule for Children
[3] ICD-10= International Classification of Diseases (10th edition)

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Li et al. (2014)	Cluster/ stratified school sample	2	92.5%	3	Waking 2+ nightly, 2+ weekly, past week	3	CSHQ[1] with additional DMS items	3	'excellent'	3 .93	20505 (49.5%)	9.0 (1.6) *5-12*	9.8%
Liu et al. (2000)	Representative/ stratified school sample	2	97.0%	3	DMS, often,	3	Key symptoms	3	NS	0 .73	1365 (60.3%)	14.6 (3.4) *12-18*	6.3%
Liu et al. (2008)[2]	Representative/ random school sample	2	97.8%	3	DMS, 3+weekly, past month	3	Key symptoms	3	α= .5[3] k= .4	2 .87	1066 (58.6%)	Median= 14.5 *12-17*	7.1%
Mak et al. (2010)	Population survey of obesity	3	84.5%	3	DMS, past 30 days	2	Key symptoms	3	NS	0 .73	28839 (49.7%)	*12-18*	14.2%
Mak et al. (2012)	Population survey of obesity	3	94.8%	3	DMS, past 30 days	2	Key symptoms	3	k= .26	2 .87	22678 (41.6%)	*12-18*	8.8%
Morioka et al. (2013)	Random school sample. Part of wider survey	3	92.2%	3	DMS, always/ often, past 30 days	3	Key symptoms	3	NS	0 .80	98867 (49.4%)	*12-18*	10.5%
Murray et al. (2012)[4]	Advertisements	2	56.0%	1	DMS, often/ frequently/ always	2	Adolescent sleep-wake scale[5]	3	α= .81	3 .73	60 (33%)	15.1 (1.8) *12-18*	8.5%

[1] CSHQ= Child Sleep Habits Questionnaire (Owens, Spirito & McGuinn, 2000)
[2] 7th and 10th grades only
[3] α = Cronbach's
[4] Control group only
[5] LeBourgeois et al. (2005)

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Nevéus et al. (1998)[1]	Single school	1	91.0%	3	"Night-time awakenings", monthly/ nightly/ weekly	2	Measure designed to study sleep problems	2	NS	0 .53	100 (49.0%)	8.7 (1.0) *7-10*	48.0%
Ohayon & Roberts (2001)[2]	Population/ stratified sample	2	78.0%	3	DSM-IV/ ICSD	3	Sleep-EVAL.	3	k= .71-.78	3 .93	794 (54.1%)	*15-18y*	8.4%
Pagel et al. (2007)	Two schools	2	47.6%	0	DMS, always/ every night	2	PSQ	3	NS	0 .47	165 (49.1%)	14.2 (1.5) *11-17*	10.3%
Pan et al. (2012)	Random school sample	2	China= 95.1% Macau = 95.7%	3	ICSD-2	3	ICSD-2	3	NS	0 .73	China= 861 (47.6%) Macau= 618 (41.9%)	China = 15.3 (1.8) Macau = 15.6 (1.8)	China = 3.7% Macau = 2.8%
Roberts et al. (2004)	Stratified school sample	2	67.0%	2	DMS, often/ every day, past 4 weeks	3	DSM-IV	3	NS	0 .67	5118 (NS)	Median= 15.0 *13-18*	14.9%
Roberts et al. (2008)[3]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interview based on DSM-IV	3	NS	0 .73	4175 (51.1%)	*11-17*	3.1%

[1] Control group only
[2] 15-18 years only
[3] Wave 1 only

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Roberts et al. (2002)	Epidemiological study	3	NS	0	DMS, almost every day, past 4 weeks	3	DSM-IV	3	α >.9[1]	3 .8	4175 (50.8%)	*11-17*	8.7%
Shang et al. (2005)	Stratified school sample	2	91.6%	3	Waking 30+ mins, 3+ weekly, past month	3	SHQ[2]	3	k= .54-.85	3 .90	1391 (52.4%)	7.4 (1.5) *4-9*	1.5%
Shur-Fen (2006)	Random school sample	2	95.3%	3	DMS, 30+ mins 3+ weekly, for 1+ month over past 6 months	3	SHQ	3	NS	0 .73	2463 (52.4%)	11.6 (2.6) *6-16*	1.4%
Singareddy et al. (2013)[3]	Population/ stratified school sample	2	70.0%	3	DMS, very often, past 2 months.	3	Key symptoms	3	NS	0 .73	666 (48.0%)	8.8 (1.7) *5-12*	21.3%
Siomos et al. (2009)	Cross-section of schools	2	99.8%	3	ICD-10	3	AIS[4]	3	α= .82	3 .93	2195 (49.1%)	15.3 (1.7) *13-18*	1.8%
Stein et al. (2001)	Paediatric clinics	1	NS	0	DMS, 30+ mins 7x weekly	2	Key symptoms	3	NS	0 .40	472 (53.6%)	*4-12*	4.0%

[1] α= Cronbach's
[2] SHQ= Sleep Habits Questionnaire (Gau, 2000)
[3] Non-self-harming group only
[4] AIS= Athens Insomnia Scale (Soldatos et al., 2000)

Table 1.10 – Summary of articles reporting the prevalence of difficulty maintaining sleep in non-clinical samples.

Authors	Quality ratings											N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Zhang et al. (2009)	Multiple schools	2	70.3%	3	DSM-IV/ ICSD	3	DSM-IV/ ICSD	3	NS	0	.73	5695 (50.3%)	9.2 (1.8) *6-13*	1.3%
Zhang et al. (2011)[1]	Epidemiological study	2	47.2%	0	DMS, 3+ weekly, past 12 months	3	Key symptoms	3	α= .87	3	.73	6447 (50.6%)	9.2 (1.8)	1.3%
Zhang et al. (2015)	Random school sample	2	76.4%	3	DMS, 3+ weekly, past 12 months	3	Key symptoms	3	NS	0	.73	3086 (52.1%)	9.7 (1.7)/ 9.0 (1.6) *7-14*	1.3%

[1] Baseline data only

There was substantial overlap between articles reporting prevalence estimates for DMS and those reporting DIS. Therefore, the methods of recruitment and assessment were very similar. In terms of the definition of DMS, some authors required a minimum frequency or duration of night-wakings, in order for DMS to be considered as present (Huang, Wang & Guilleminault, 2010; Kirmil-Gray, Eagleston, Gibson & Thoresen, 1984; Shur-Fen, 2006), whereas others described DMS as being restless sleep, or difficulty in returning to sleep once awake, without using operationalised terms of how this might present clinically (Amaral et al., 2013; Liu, Zhao, Jia & Buysse, 2008; Roberts et al., 2008b; Roberts, Roberts & Chen, 2002; Singareddy et al., 2013).

The article by Li and colleagues (2014) was the only publication to focus solely on DMS. The aim of the study was to investigate the impact of psychosocial factors, such as demographic variables, health and the sleeping environment, on whether a child experienced frequent night-time awakenings, which was the term employed by the authors. The authors' definition was therefore more detailed, and required participants to wake more than twice each night, or awaken once during the night for a duration of longer than 30 minutes, for a minimum of two nights per week during the past four weeks, to be classified as experiencing DMS. A prevalence rate of 9.8% was observed in their sample of over 20,000 children under the age of 12 years, and this was found to be influenced by health problems, poor psychological and emotional functioning, and poor sleep hygiene. This article was given high scores on the quality framework due to the clear definition of the construct, high participant uptake rate, and valid assessment of DMS. Articles which received lower scores on the quality framework were predominantly those that

either did not report a participant uptake rate, or whose uptake rate was relatively low in comparison to the other papers (Barclay et al., 2015; Gehrman et al., 2011; Kirmil-Gray et al., 1984; Lehmkuhl, Wiater, Mitschke & Fricke-Oerkermann, 2008; Pagel, Forister & Kwiatkowki, 2007; Stein et al., 2001). The articles were entered into the pooled meta-analysis to give overall prevalence estimates (Figures 1.8 and 1.9).

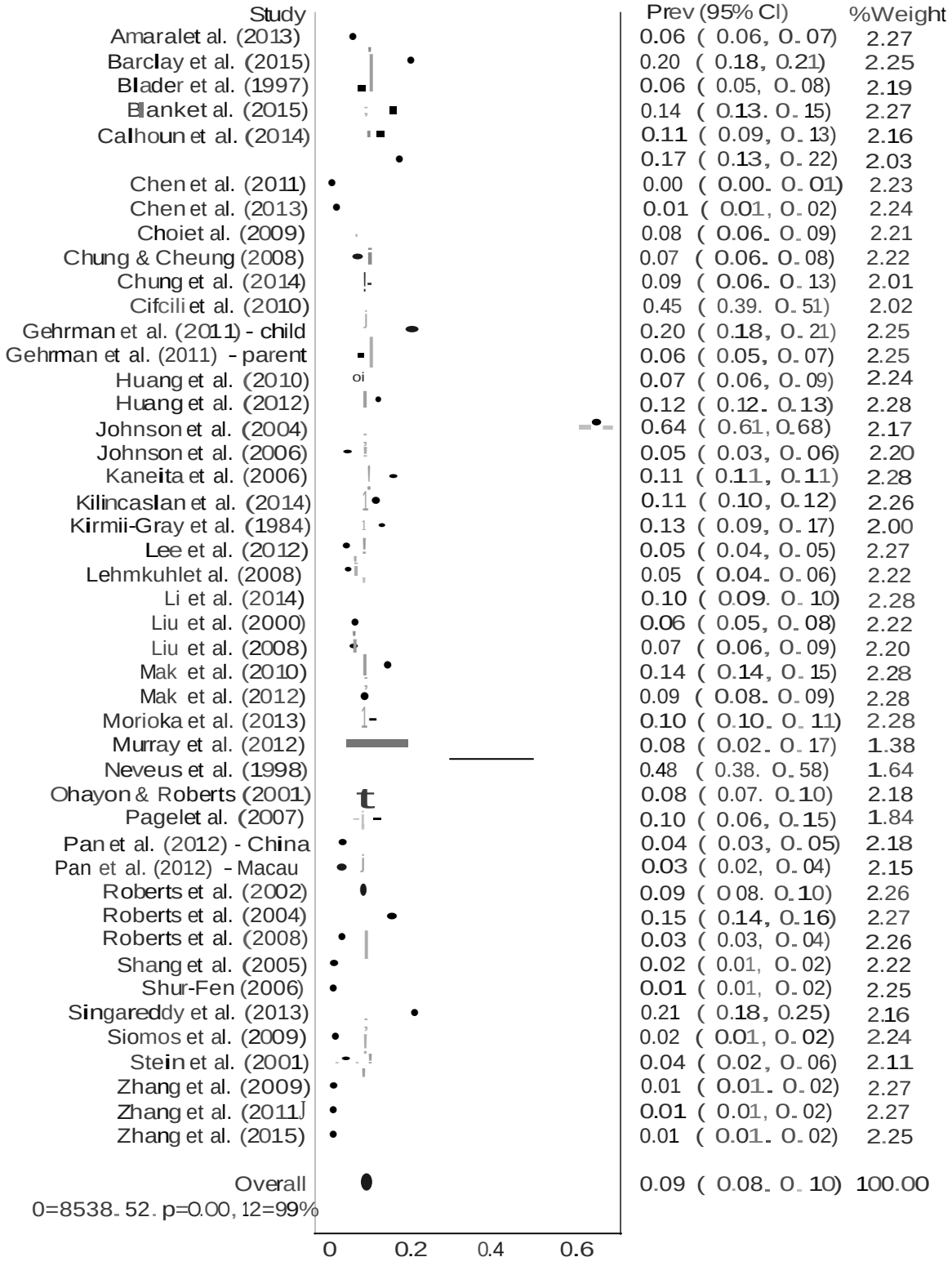

Figure 1.8- Pooled estimates for the prevalence of difficulty maintaining sleep, using a random-effects model.

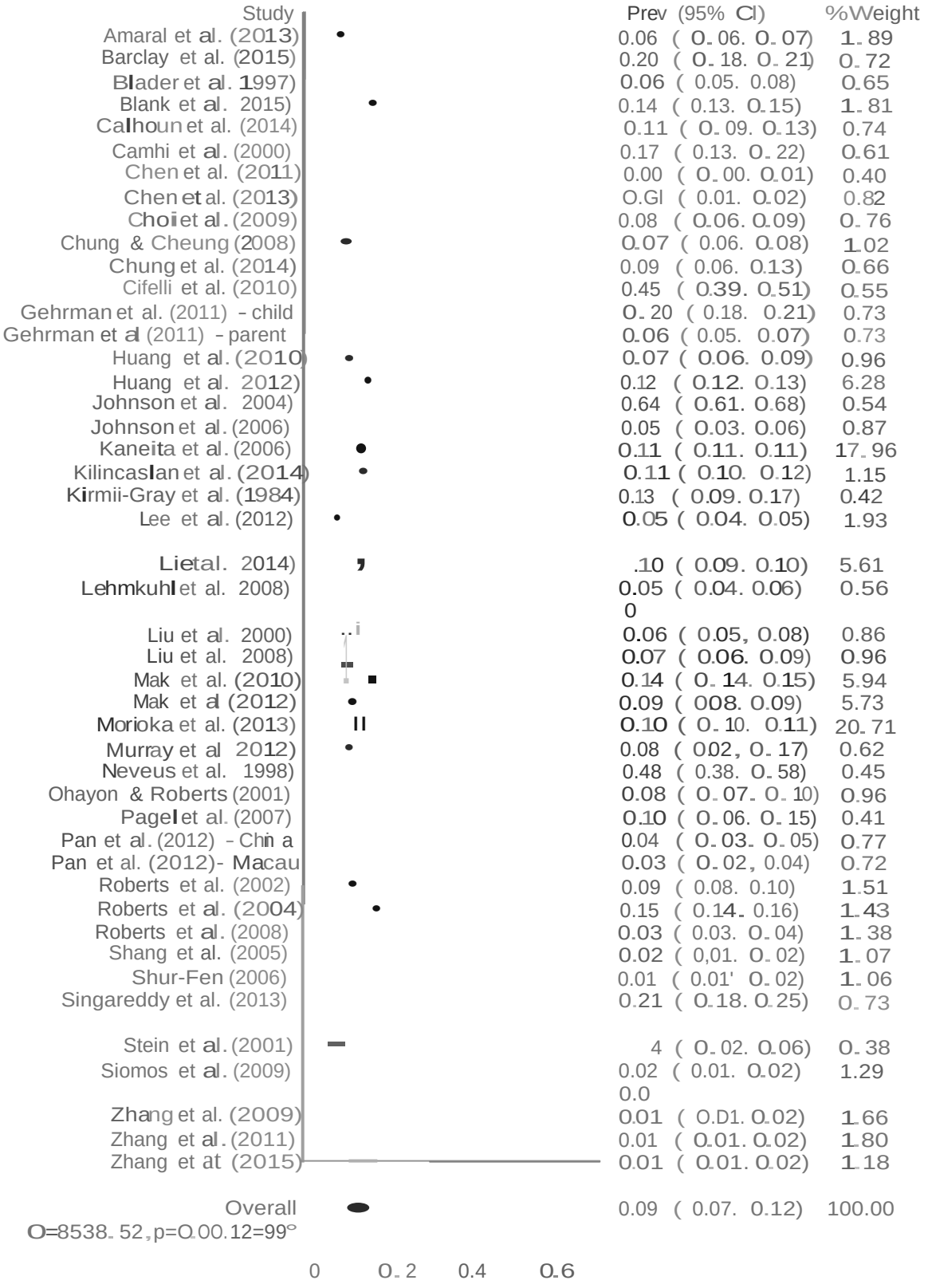

Figure 1.9- Pooled estimates for the prevalence of difficulty maintaining sleep, using a quality-effects model.

The results of the pooled estimates suggested that the prevalence of DMS is 8.8% (CI= 7.5-10.2) for the random-effects model, and 9.4% (CI= 6.8-12.4) for the quality-effects model. These are summarised in Figures 1.8 and 1.9.

As with the DIS analysis, the Nevéus et al. (1998) article reported a prevalence that appeared to be anomalous, and again it is likely that because the authors chose a more liberal frequency of monthly occurrences of DMS, a greater number of children in their sample met the criteria. Other articles that stood out as being inconsistent with the other prevalence estimates, were those of Çifçili et al. (2010) and Johnson et al. (2004). These authors also used slightly different definitions of DMS, in that Çifçili and colleagues enquired about the presence of DMS and did not stipulate a frequency of symptoms, or duration of chronicity. Johnson and colleagues (2004) combined DMS with early morning awakening (EMA), and so this most likely inflated the number of participants who responded positively to that item. Removal of these three articles from the pooled prevalence estimates, yielded new estimates of 7.1% (CI= 6.0-8.3) and 8.9% (CI= 6.6-11.6) from the random- and quality-effects models respectively. This is a more conservative estimate of DMS, and is likely more in keeping with what would be considered a problem worthy of clinical investigation; however, the overlap between the confidence intervals suggests that removal of the three papers does not significantly change the prevalence estimates, and the original estimates were therefore retained.

Early morning awakening. Twenty-six articles were identified that reported a prevalence figure for early morning wakening (EMA), after three had been removed for including non-unique samples (Liu, 2004; Liu & Zhou, 2002; Roberts et

al., 2006) and one for failing to score on the three key criteria (Lazaratou et al., 2005). These are summarised in Table 1.11. Reported prevalence estimates for the remaining articles ranged from 0.2-17.3% (Blanket al., 2015; Gehrman et al., 2011).

Table 1.11 – Summary of articles reporting the prevalence of early morning awakening in non-clinical samples.

Authors	Quality ratings											N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Amaral et al. (2013)	Multiple schools	2	82.1%	3	DSM-IV[1]	3	DSM-IV	3	NS	0	.73	6919 (47.0%)	*12-18*	8.2%
Barclay et al. (2015)[2]	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA[3]	3	NS	0	.47	2789 (46.0%)	Mode= 8.3 *8-17*	4.9%
Blank et al. (2015)	National survey	3	82.9%	3	DSM-IV	2	DSM-IV	3	NS	0	.73	6483 (48.6%)	*13-18*	17.3%
Choi et al. (2009)[4]	Random/ stratified school sample	2	86.7%	3	EMA[5], 3+ weekly	2	Key symptom	3	NS	0	.67	1176 (50.0%)	16.7 (1.0)	6.3%
Chung & Cheung (2008)[6]	Multiple schools	2	86.0%	3	EMA, 3+ weekly, past 3 months	3	Sleep Quality Index[7]	3	r= .61[8]	2	.87	1335 (NS)	*12-16*	10.4%
Chung et al. (2014)	Stratified school sample	2	72.0%	3	DSM-IV	3	Insomnia Interview Schedule[9]	3	NS	0	.73	290 (43.8%)	14.4 (1.4) *12-18*	6.6%
Gehrman et al. (2011)	Longitudinal twin study	1	NS	0	DSM-III-R	3	CAPA	3	NS	0	.47	2824 (46.3%)	12.0 (2.6) *8-16*	Child = 4.9%, Parent = 0.2%

[1] DSM= Diagnostic and Statistical Manual
[2] Wave 1 only
[3] CAPA= Child and Adolescent Psychiatric Assessment (Angold et al., 1995)
[4] Control group only
[5] EMA= Early morning awakening
[6] 12-16 years only
[7] Urponen et al., 1991
[8] r= test-retest reliability/ inter-rater reliability
[9] Morin (1993)

Table 1.11 – Summary of articles reporting the prevalence of early morning awakening in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Huang et al. (2012)	Stratified school sample, part of wider survey	3	NS	0	EMA, past 30 days	2	Key symptoms	3	k= .26[1]	1	.60	33692 (44.9%)	*13-15*	9.3%
Huang et al. (2010)	Random school sample	2	95.1%	3	EMA	1	PSQ	3	r= .64[1]	2	.73	1906 (37.7%)	*12-18*	15.1%
Junker et al. (2014)[2]	Population sample for non-sleep study	3	88.0%	3	EMA, often/ almost every night, past month	3	Key symptom	3	NS	0	.73	4325 (NS)	*13-16*	4.0%
Kaneita et al. (2006)	Multiple schools	2	64.8%	2	EMA, often/ always, past month	3	Key symptom	3	NS	0	.67	102451 (54.7%)	*12-18*	5.5%
Lee et al. (2012)	School sample	2	84.1%	3	ICD-10[3]	3	ICD-10	3	NS	0	.73	7172 (37.8%)	16.8 (1.1) *12-17*	1.4%
Liu et al. (2000)	Stratified school sample	2	97.0%	3	EMA, often, past month	3	Key symptom	3	NS	0	.73	1365 (60.3%)	14.6 (3.4) *12-18*	2.1%
Liu et al. (2008)[4]	Representative/ random school samples	2	97.8%	3	EMA, 3+ weekly, past month	3	Key symptom	3	α= .5 k= .4	2	.87	1066 (58.6%)	Median= 14.5 *12-17*	3.4%
Mak et al. (2010)	Total population survey of obesity	3	84.5%	3	EMA, past 30 days	2	Key symptom	3	NS	0	.73	28839 (49.7%)	*12-18*	10.3%

[1] k or α= test-retest reliability/ inter-rater reliability
[2] Under 16 years only
[3] ICD-10= International Classification of Diseases (10th edition)
[4] 7th and 10th grades only

Table 1.11 – Summary of articles reporting the prevalence of early morning awakening in non-clinical samples.

Authors	Quality ratings											N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Mak et al. (2012)	Total population survey of obesity	3	94.8%	3	EMA, past 30 days	2	Key symptom	3	k= .26	2	.87	22678 (41.6%)	*12-18*	7.2%
Morioka et al. (2013)	Random school sample, part of wider survey	3	92.2%	3	EMA, always/ often, past 30 days	3	Key symptom	3	NS	0	.80	98867 (49.4%)	*12-18*	5.1%
Ohayon & Roberts (2001)[1]	Population/ stratified sample	2	78.0%	3	DSM-IV/ ICSD[2]	3	Sleep-EVAL	3	k= .78-.71	3	.93	794 (54.1%)	*15-18*	10.5%
Pan et al. (2012)	Random school sample	2	China= 95.1% Macau =95.7%	3	ICSD-2	3	ICSD-2	3	NS	0	.73	China= 861 (47.6%) Macau= 618 (41.9%)	China = 15.3 (1.8) Macau = 15.6 (1.8)	China= 10.5% Macau = 5.8%
Roberts et al. (2004)	Stratified school sample	2	67.0%	2	EMA, often/ daily, past 4 weeks	3	DSM-IV	3	NS	0	.67	5118 (NS)	Median= 15.0 *13-18*	15.1%
Roberts et al. (2008)[3]	Epidemiological study	3	66.0%	2	DSM-IV	3	Interviews based on DSM-IV	3	NS	0	.73	4175 (51.1%)	*11-17*	3.2%
Roberts et al. (2002)	Epidemiological study	3	NS	0	EMA, almost/ daily, past 4 weeks	3	DSM-IV	3	α >.9	3	.8	4175 (50.8%)	*11-17*	3.3%

[1] 15-18 years only
[2] ICSD= International Classification of Sleep Disorders
[3] Wave 1 only

Table 1.11 – Summary of articles reporting the prevalence of early morning awakening in non-clinical samples.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Siomos et al. (2009)	Cross-section of schools	2	99.8%	3	ICD-10	3	AIS[1]	3	α= .82	3	.93	2195 (49.1%)	15.3 (1.7) *13-18*	2.2%
Zhang et al. (2009)	Multiple schools	2	70.3%	3	DSM IV/ ICSD-1	3	DSM IV/ ICSD-1	3	NS	0	.73	5695 (50.3%)	9.2 (1.8) *6-13*	1.2%
Zhang et al. (2011)[2]	Epidemiological study of sleep problems	2	47.2%	0	EMA, 3+ weekly, past 12 months	3	Key symptom	3	α= .87	3	.73	6447 (50.6%)	9.2 (1.8)	1.2%
Zhang et al. (2015)	Random school sample	2	76.4%	3	EMA, 3+ weekly, past 12 months	3	Key symptoms	3	NS	0	.73	3086 (52.1%)	9.7 (1.7)/ 9.0 (1.6) *7-14*	1.4%

[1] AIS= Athens Insomnia Scale (Soldatos et al., 2000)
[2] Baseline data only

It was surprising to observe that authors did not typically give a cut-off for the time at which an awakening would be considered early in the articles describing prevalence rates of EMA. This allows for some interpretation on the respondent's behalf, as to how early their child would have to wake for it to be perceived as problematic. However, a frequency of three times a week was given more consistently in the present collection of articles than it had been in the articles describing the prevalence of insomnia, DIS or DMS. EMA was never reported in the absence of the other forms of insomnia, and was typically the lowest prevalence of the three symptoms.

As with the DIS and DMS comparisons, articles that received the highest ratings for quality, had representative sample recruitment strategies and high participant response rates (Ohayon & Roberts, 2001; Siomos et al., 2009), in comparison to those who employed non-representative samples or had not reported an adequate uptake rate (Barclay et al., 2015; Gehrman et al., 2011). All of the articles listed in Table 1.11 were then combined in the pooled prevalence analyses (Figures 1.10 and 1.11).

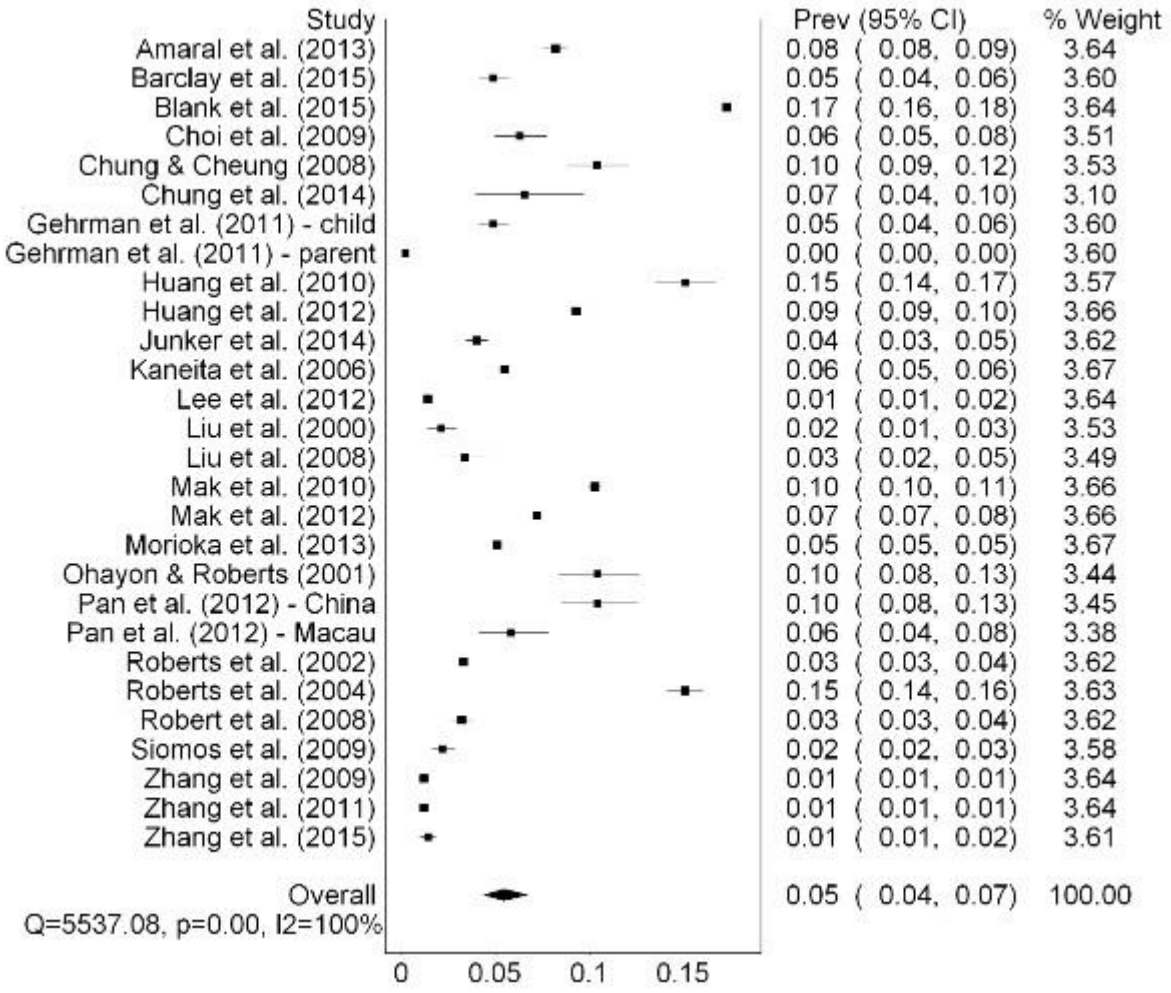

Figure 1.10 - Pooled estimates for the prevalence of early morning awakening, using a random-effects model.

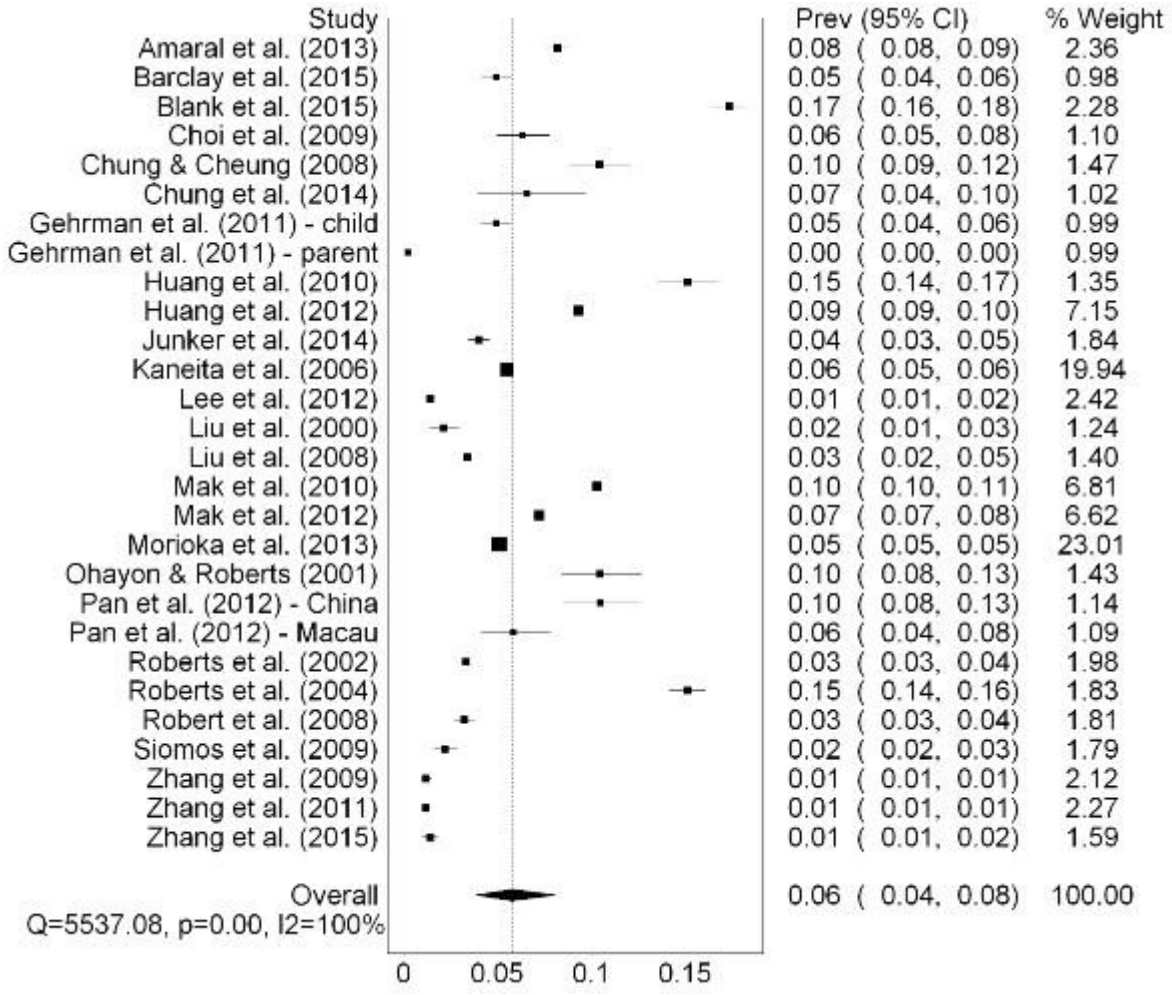

Figure 1.11 - Pooled estimates for the prevalence of early morning awakening, using a quality-effects model.

The pooled estimates for the prevalence of EMA were 5.5% (CI= 4.4-6.7) and 5.6% (CI= 3.8-8.1) for the random- and quality-effects models respectively. These results suggest that, of the three symptoms of insomnia, EMA is the least common. Alternatively, it may be that parents perceive their child waking early in the morning to be less problematic, than when they awake during the night or have difficulty falling asleep at bedtime. They therefore may not consider this to be worthy of report in a questionnaire enquiring about sleep problems. Furthermore, parents

may not be aware of their child waking early in the morning if the child remains in their room, and does not alert them to the fact that they are awake.

It should also be noted that of the three symptoms, EMA was reported by the fewest number of articles. Furthermore, papers reporting the prevalence of insomnia overall would not always include EMA in their criteria, but would more frequently include DIS and DMS (Çifçili et al., 2010; Fernandez-Mendoza et al., 2014; Kilincaslan et al., 2014; Murray, Murphy, Palermo & Clarke, 2012; Patten et al., 2000; Roane & Taylor, 2008). It may be that EMA is therefore overlooked both empirically and clinically, or it could be that, due to its reduced prevalence in comparison to DIS and DMS, it is not given as much attention in the applied or empirical arenas.

1.3.2 Prevalence of Insomnia in Mental Health Conditions

Anxiety disorders. Three articles which included a sample of children and/ or adolescents with a diagnosis of anxiety or generalised anxiety disorder were grouped together, and have been summarised in Table 1.12. Reported prevalence rates ranged from 30.4-76.5% (Johnson et al., 2006a; Ohayon, Roberts, Zulley, Smirne & Priest, 2000).

Table 1.12 – Summary of articles reporting the prevalence of insomnia in anxiety disorders.

Authors	Quality ratings											N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability		Total			
Alfano et al. (2007)[1]	Medical trial	1	NS	0	DIS/ DMS[2]	1	Key symptoms	3	NS	0	.33	128 (50.8%)	10.7 *6-17*	56.9%
Johnson et al. (2006)	Health insurance database	2	71.2%	3	DIS/ DMS/ NRS[3], 4+ weekly, w. daytime impact, 1 month in lifetime	3	DSM/ ICSD[4]	3	NS	0	.73	1014 (49.7%)	14.4 *13-16*	30.4%
Ohayon et al. (2000)	Stratified/ probability sample	2	80.8%	3	DSM-IV/ ICSD	3	DSM-IV/ ICSD	3	k= .70-.78[5]	3	.93	1125 (54.1%)	*15-18*	76.5%

[1] Baseline only
[2] DIS= Difficulty initiating sleep; DMS= Difficulty maintaining sleep
[3] NRS= Non-restorative sleep
[4] DSM= Diagnostic and Statistical Manual; ICSD= International Classification of Sleep Disorders
[5] k= inter-rater reliability

Each article was entered into a random- and quality-effects model to determine the pooled prevalence statistics.

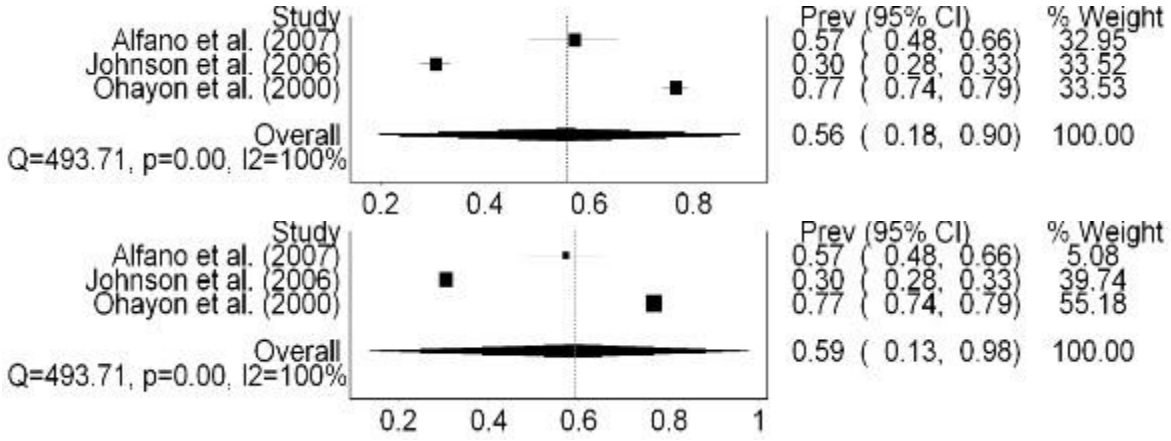

Figure 1.12 - Pooled estimates for the prevalence on insomnia in anxiety disorders, using random-effects (upper panel) and quality-effects (lower panel) models.

The pooled prevalence statistics for the rates of insomnia in children with a comorbid anxiety disorder were 55.5% (CI= 17.7-90.5) for the random-effects model, and 59.0% (CI= 13.4-97.6) for the quality-effects model. As with the rates of insomnia in the wider population, there was an extensive level of heterogeneity within the data. This is reflected in the very large confidence intervals for both models. As with previous analyses, this is most likely down to the authors varying definitions of insomnia. For example, Alfano, Ginsburg and Newman Kingery (2007) required only the presence of DIS or DMS for participants to score positively for insomnia, whereas Johnson et al. (2006a) applied more stringent criteria, and only included participants who were also experiencing daytime distress or impairment. Furthermore, the article by Ohayon et al. (2000) reported “insomnia symptoms” in 76.5% of their adolescent sample who met criteria for an anxiety disorder, however it is unclear whether these insomnia symptoms equated to meeting diagnostic

criteria, and whether several types of anxiety were included under the term ‘anxiety disorder'.

Depression. Several articles identified within the initial search also included samples of children and adolescents with a diagnosis of depression. For the purpose of the present analysis, those with a diagnosis of major depressive disorder and those with depression have been combined. Six articles were identified, yet two were removed due to failing to score on one of the key items of definition, recruitment or assessment (Strober, Green & Carlson, 1981; Yorbik, Birmaher, Axelson, Williamson & Ryan, 2004). The remaining articles are summarised in Table 1.13. Prevalence estimates ranged from 54.5-72.7% (Murray et al., 2012; Roberts, Lewinsohn, & Seeley, 1995).

Table 1.13 – Summary of articles reporting the prevalence of insomnia in depression.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Liu et al. (2007)	Multiple psychiatric facilities	2	NS	0	DIS/ DMS/ EMA[1]/ poor sleep quality/ sleeping less, nearly every day, 2+ weeks during past month	3	ISCA[2], based on DSM[3]	3	NS	0 .53	553 (55.0%)	11.7 (2.0) *7.0-14.9*	64.0%
Murray et al. (2012)	Sample from ongoing study, or prescription database	2	21.0%	0	DIS/ DMS, often/ frequently/ always	2	Key symptoms	3	α= .81[4]	3 .83	46 (33.0%)	15.1 (1.8) *12-18*	72.7%
Roberts & Duong (2013)[5]	Health insurance databases	2	66.0%	2	DIS/ DMS/ EMA/ NRS[6], past 4 weeks	3	DSM/ ICSD[7]	3	NS	0 .67	Total incl. dep. group= 4175 (51.0%)	11.7 (2.0) *7-14.9*	56.0%
Roberts et al. (1995)[8]	Random school sample	3	61.0%	2	DIS	1	K-SADS[9]	3	k >.8[10]	3 .73	44 (46.3%)	16.6 (1.2)	54.5% (DIS only)

[1] DIS= Difficulty initiating sleep; DMS= Difficulty maintaining sleep; EMA= Early morning awakening
[2] ISCA= Interview schedule for children and adolescents (Sherrill & Kovacs, 2000)
[3] DSM= Diagnostic and Statistical Manual
[4] α= internal consistency
[5] Baseline only
[6] NRS= Non-restorative sleep
[7] ICSD= International Classification of Sleep Disorders
[8] Time 1 only
[9] K-SADS= Schedule for affective disorder and schizophrenia for school-age children: Kiddie-SADS (Chambers et al., 1985)
[10] k= Cohen's k

Articles were then entered into the random- and quality effects models, to give overall prevalence estimates.

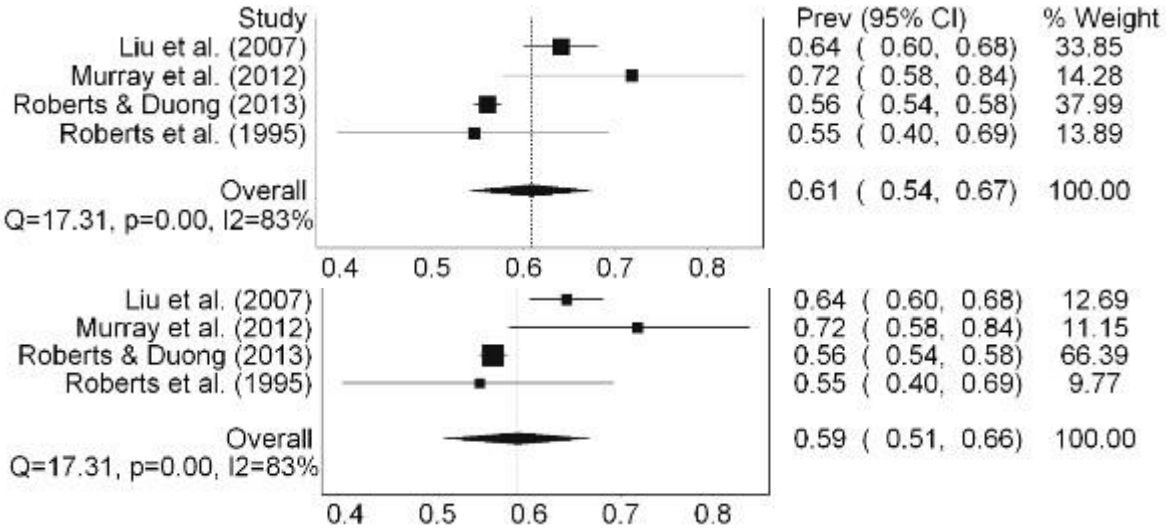

Figure 1.13 - Pooled estimates for the prevalence of insomnia in depression, using random-effects (upper panel) and quality-effects (lower panel) models.

The pooled estimates for the prevalence of insomnia in children with comorbid experiences of depression were 60.8% (CI= 54.0-67.4) and 58.7% (CI= 50.7-66.5) for the random- and quality-effects models respectively. Although the collection of papers was slightly more homogenous than in previous analyses, the observed heterogeneity was still significant. One explanation for this could be that the Roberts et al. (1995) paper reported individual symptoms of insomnia, rather than an overall prevalence, and so the prevalence estimate for DIS was chosen to enter into the meta-analysis. The remaining articles all reported a prevalence estimate for insomnia, defined as being frequent symptoms of insomnia, with the additional criterion of added daytime impairment in the Roberts & Duong (2013) article, and 'clinical significance' in the paper by Liu et al. (2007; pg. 85).

Another key difference between the articles is the recruitment strategy. Whilst the two papers published by Roberts and colleagues (1995; 2013) recruited

a typically-developing sample, and identified participants experiencing symptoms of depression within that group, Liu et al. (2007) and Murray et al. (2012) recruited participants through existing populations of psychiatric patients. Unfortunately, the Liu et al. (2007) article did not report a participant uptake rate, and the participant uptake rate reported in the Murray et al. (2012) sample was very low. Thus, it may be that a response bias has occurred in these two samples, and that individuals experiencing sleep problems alongside a mood disorder were more likely to participate. This may be especially pertinent when considering the clinical characteristics of depression, and the heightened risk of apathy in this group. This may provide some explanation for the higher prevalence of insomnia reported in these two articles, in comparison to those by Roberts and colleagues (1995; 2013).

1.3.3 Prevalence of Insomnia in Physical Health Conditions

As described previously, insomnia has been suggested as being more prevalent in physical health conditions in addition to mental health diagnoses. The articles that had been identified as including participants with physical health conditions, were grouped according to their diagnosis, for meta-analysis of the reported prevalence rates for each condition. However, after exclusion of articles failing to score positively on the three key criteria of definition and assessment of insomnia, and adequate sample recruitment, the only diagnosis to retain a collection of more than one article was that of chronic pain.

Chronic pain. Of the articles including a sample of participants with physical health conditions, chronic pain featured in four articles. These are summarised in Table 1.14.

Table 1.14 – Summary of articles reporting the prevalence of insomnia in chronic pain.

Authors	Quality ratings										N (% Male)	Mean age in years (SD) *range*	Prev.
	Sample recruitment		Response rate		Definition		Assessment		Reliability	Total			
Kanstrup et al. (2014)	Specialist centre	1	NS	0	DSM-IV[1]	3	ISI[2]	3	α= .88[3]	3 .67	154 (24.7%)	14.6 (2.0) *10-18*	51.7%
Meltzer et al. (2005)	Outpatient clinic	1	96.0%	3	DMS[4] of 2+ wakings, over past fortnight	3	Sleep Habits Survey[5] and telephone interview	3	α= .7-.75	3 .87	26 (0%)	15.5 *12-17*	62.5%
Palermo et al. (2007)	Pain clinic	1	86.0%	3	DIS of 30+ mins/ 3+ nightly wakings, 60%+ of time	2	Actigraphy, Adolescent Sleep Wake Scale[6]	3	NS	0 .60	20 (25.0%)	15.1 (1.4) *12-17*	55.0%
Palermo et al. (2011)	Pain clinic	1	62.1%	2	DIS/ DMS, frequently/ quite often/ always	2	Key symptoms	3	α= .70	3 .73	59 (29.0%)	15.0 (1.7) *12-18*	54.2%

[1] DSM= Diagnostic and Statistical Manual
[2] ISI= Insomnia Severity Index (Morin, 1993)
[3] α= internal consistency/ reliability
[4] DIS= Difficulty initiating sleep; DMS= Difficulty maintaining sleep
[5] Wolfson and Carskadon (1998)
[6] LeBourgeois et al. (2005)

The reported prevalence rates were between 51.7-62.5% (Kanstrup, Holmström, Ringström & Wicksell, 2014; Meltzer, Logan & Mindell, 2005). The four articles were then entered into the pooled prevalence analyses (Figure 1.14).

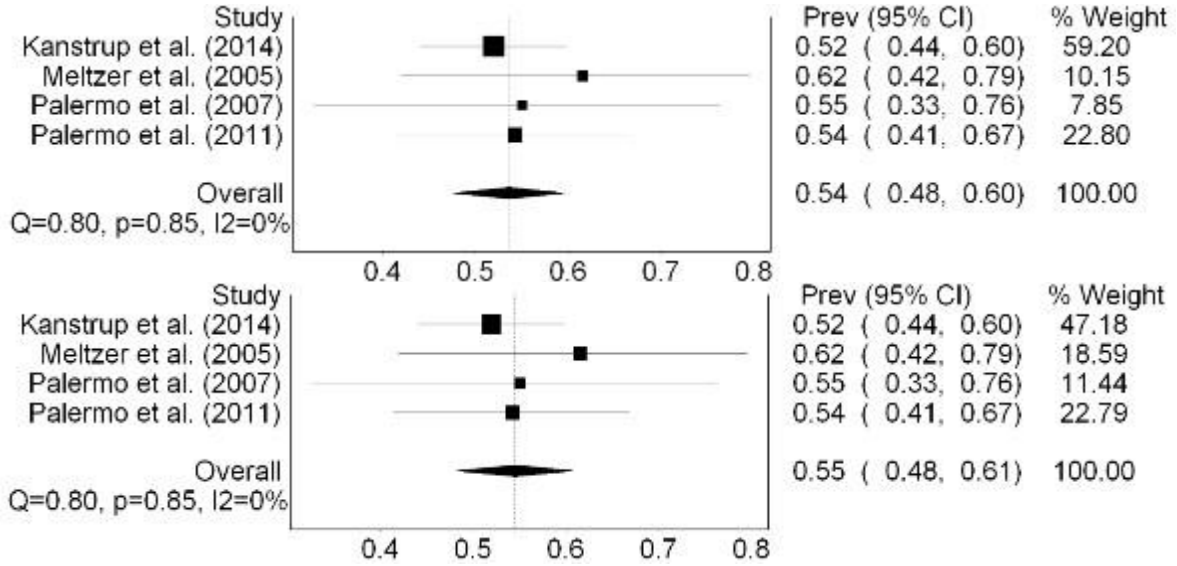

Figure 1.14 - Pooled estimates for the prevalence of insomnia in chronic pain, using random-effects (upper panel) and quality-effects (lower panel) models.

The pooled estimate for the prevalence of insomnia in children experiencing chronic pain was 53.6% in the random-effects model (CI= 47.5-59.7), and 54.5% in the quality-effects model (CI= 48.1-60.8). No significant heterogeneity was observed between the articles, which is likely attributable to the similarities in the source from which the samples were recruited, and the recommended chronicity of 3 months of experienced chronic pain. Furthermore, both articles authored by Palermo and colleagues (Palermo, Toliver-Sokol, Fonareva & Koh, 2007; Palermo et al., 2011) included the frequent occurrence of DIS or DMS as being indicative of insomnia. Kanstrup et al. (2014) used a cut-off of nine on the Insomnia Severity Index (ISI: Morin, 1993) which is comparable to a moderate level of clinical insomnia. Each of these definitions are of a similar level of stringency. Meltzer and colleagues (2005) stipulated that DMS needed to be present during two nights of a

fortnight, which is a less conservative definition, reflected in the slightly higher reported prevalence rates.

1.3.4 Summary

The final aim of the meta-analysis was to compare the prevalence of insomnia, in clinical and non-clinical populations. Figure 1.15 summarises the prevalence estimates according to the random- and quality-effects models, with 95% confidence intervals, and demonstrates the increased prevalence of insomnia in the three clinical populations. Odds ratios for the quality effects data ranged from 4.8 to 5.9 in the three clinical groups (Table 1.15), again, suggesting that children and adolescents with a physical or mental health problem have a higher chance of presenting with insomnia.

Table 1.15 - Odds ratios for presence of insomnia in each clinical sample in comparison to the non-clinical estimates

	Odds Ratio (95% Confidence Intervals)	
	Random Effects	Quality Effects
Anxiety	5.11 (2.72-9.60)	5.77 (3.07-10.84)
Depression	6.88 (3.64-13.00)	5.86 (3.11-11.02)
Chronic Pain	4.62 (2.47-8.67)	4.79 (2.56-8.99)

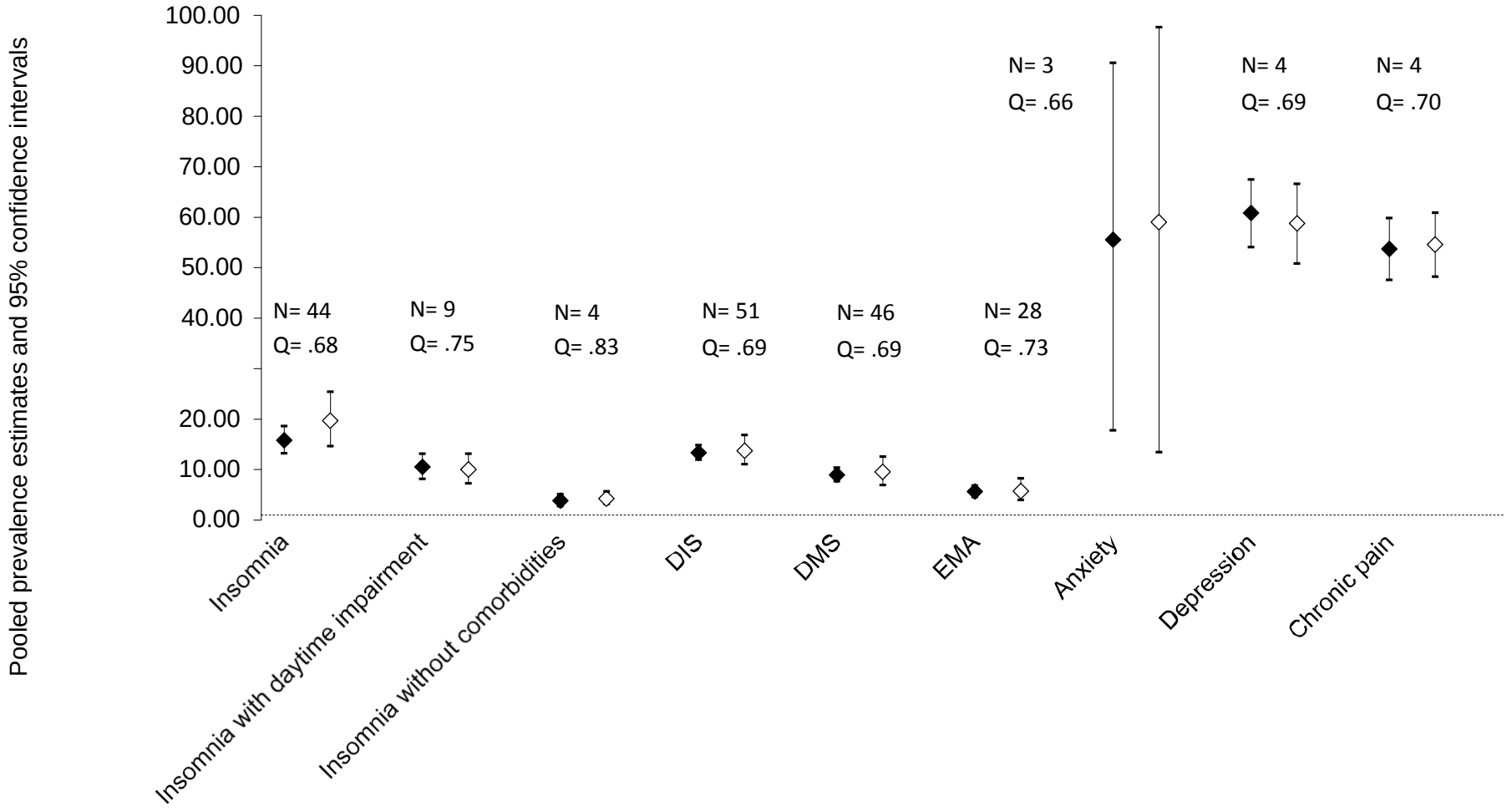

Figure 1.15 - Pooled prevalence estimates and 95% confidence intervals for each definition of insomnia in the non-clinical sample, and for each clinical sample. N= Number of articles in analysis; Q= Mean quality weighting; ◆= Random effects; ◇= Quality effects.

1.4 Discussion

The aim of the current systematic review and meta-analysis was to delineate the prevalence of insomnia and its components in non-clinical child and adolescent populations, and compare this to the prevalence of insomnia in physical and mental health conditions commonly associated with insomnia. This is the first study to undertake an extensive search of the empirical literature, in order to synthesise the range of reported prevalence rates in this age-group. In addition, careful evaluation of the varying definitions of the construct of insomnia, and the methodology employed to assess its presence has been undertaken.

The overall findings of the meta-analyses suggest that the prevalence of insomnia in children and adolescents is between 16 and 20%. The prevalence was comparable between adolescents and children, with the prevalence of insomnia estimated at 18-21% in adolescent samples and 19-20% in child samples under 12 years of age. With regards to the prevalence of each symptom of insomnia, DIS was the most frequently reported with a prevalence of 13-14%, followed by DMS which had a reported prevalence of 9%. EMA was the least common symptom of insomnia and occurred in 6% the participants included here. The observation that DIS was the most prevalent of the symptoms of insomnia suggests that this would be the most effective focus of intervention. This is consistent with the most common current treatment options available, which are typically designed to encourage night-time settling. These include behavioural therapies to develop helpful bedtime routines and reduce unhelpful bedtime associations, and pharmacological treatments, either prescribed by a medical professional or purchased by parents over the counter (Taylor & Roane, 2010).

Despite the occurrence of daytime impairment being one of the key criteria in recent DSM and ICSD publications, this was reported infrequently in the research literature. Those that did include this element in their definition of insomnia contributed to an overall prevalence of 10%. This prevalence estimate is considerably lower than when insomnia is based on the presence of symptoms alone, and implies that there are a number of children and adolescents who experience the sleeplessness symptoms of insomnia, but do not present with subsequent daytime consequences. This raises the question of whether this element of the insomnia diagnosis is clinically relevant. If a child experiences features of sleeplessness, is this less worthy of intervention when there is no impact on their functioning the following day? If we consider sleeplessness without any negative consequences to be natural variability in children's sleep patterns, rather than indicative of a clinical problem, then a great deal of the empirical literature may not be based on insomnia at all, but on typical variance within the population. Alternatively, it may be that these children present with sleep problems at a sub-clinical level that do not cause short-term impairment to daytime functioning, and are therefore overlooked by parents and care-givers, yet they may still have significant longitudinal consequences on long-term development (Jan et al., 2010).

Another important aspect to consider is the person for whom the sleeplessness is problematic. Often, sleep measures or diaries are completed by the parent or care-giver, and their perception of their child's sleep may differ to the child's own (Fricke-Oerkermann et al., 2007; Short et al., 2013). Although several studies made use of physiological measures, such as polysomnography or actigraphy, these were typically not employed to determine whether the participant

was displaying signs of insomnia. Instead insomnia was assessed using questionnaire items based on the three key symptoms stipulated in the diagnostic criteria. Another feature of the criteria which was infrequently considered was the opportunity and environment for adequate sleep to be achieved, and in the DSM, the exclusion of alternative diagnoses that may better explain the presence of sleeplessness. There are therefore considerable improvements that need to be made in the empirical fields, when considering what is actually meant by the construct of insomnia. Without clear and agreed definitions of the disorder, empirical findings cannot be reliably generalised to clinical practice.

The series of articles by Roberts and colleagues included some of the few studies that reported multiple prevalence data, dependent on the rigour of the definition (Roberts & Duong, 2013; Roberts et al., 2008a). For example, the authors reported a prevalence for the presence of any symptom of insomnia, another for insomnia with daytime impairment, and finally, insomnia with daytime impairment in the absence of any other comorbidities, such as substance use or mental health diagnoses. These were the most thorough definitions of insomnia of the retrieved articles, and demonstrate clearly how the definition of insomnia contributes heavily to the observed prevalence. Yet, it was interesting to note that none of the articles used definitions specific to insomnia in childhood, as suggested by Mindell et al., (2006) and Glaze et al., (2002), nor was it commonplace to explore the importance of bedtime routines and associations, fears around bedtime, or the presence of appropriate sleep hygiene and sleeping environment, in order to discount other possible explanations for poor sleep in this population.

One strength of the literature is the extent to which authors attempted to gain large, representative participant samples. Although the focus of the vast majority of papers was beyond identifying prevalence rates, the manner in which participants were recruited and the high response rates were valuable when extracting meaningful prevalence data. However, this often led to a trade-off in terms of the assessment of insomnia. Many of the studies measured rates of insomnia as part of a wider screening of child and adolescent wellbeing. Whilst this reduced considerably the likelihood of a response bias distorting the prevalence data, it also meant that insomnia was a very small part of the dataset of interest, and so was often poorly operationalised or assessed with a single item. As a result, larger epidemiological studies tended to overinflate prevalence estimates, in comparison to smaller studies that had been more conservative in what they considered to meet diagnostic criteria.

When considering the prevalence estimates for children in the clinical groups, it could be seen that there was a higher proportion of participants experiencing insomnia across the mental and physical health conditions. Of the mental health diagnoses, those with signs of depressive disorder reported a prevalence of insomnia of 59-63% (OR= 5.9-6.9). In comparison, 56-59% of those with anxiety also had signs of insomnia (OR= 5.1-5.8). Both of these prevalence estimates are considerably higher than that observed in the non-clinical population. Children who were included in papers reporting the prevalence of insomnia in chronic pain conditions demonstrated a prevalence of 54-55% (OR= 4.6-4.8). It can be seen that across the clinical groups, the prevalence of insomnia was much greater than in the non-clinical samples.

These findings should be interpreted with some caution, as the number of papers from which to derive prevalence estimates was relatively small, and the level of heterogeneity was high. In particular, the confidence intervals for the anxiety group would imply that the prevalence estimate could be anywhere between 13% and 97%. This may be due to variations in the authors' definition or assessment of insomnia, or the types of anxiety disorder included within the sample. For example, Ohayon et al. (2000) reported the prevalence of insomnia symptoms in children and adolescents with anxiety disorders, compared to Alfano et al. (2007), and Johnson et al. (2006a) who reported prevalence rates in participants with a generalised anxiety disorder only. This would suggest that the Ohayon et al. paper may be more inclusive, and therefore the higher prevalence estimate is to be expected. Synthesis of the prevalence estimates in clinical samples therefore adds an additional source of variability, as differences can occur not only in the definition and assessment of insomnia, but also the clinical disorder of interest. Future research should look to clarify the multiple factors surrounding mental and physical health problems that could contribute to the development of insomnia, or the elements of insomnia that influence the manifestation of physical and mental health problems.

In summary, the findings of the meta-analyses suggest that insomnia is prevalent in 4-20% of the non-clinical child and adolescent population, dependent on the rigour of the chosen criteria. Furthermore, children and adolescents who have comorbid mental or physical health problems are considerably more likely to present with insomnia, whatever the direction of causality. Authors of future research should seek to define their construct of insomnia clearly, and ensure that if they are basing their definition on diagnostic criteria, they include these elements

in their assessment. Attention should also be given to the features of insomnia that are particular to this age group, and the methodological flaws associated with such. For example, including items on factors, such as use of technology before bed, a noisy or bright sleeping environment, or unhelpful bedtime routines or associations, may influence whether a participant is incorrectly classified as experiencing insomnia, therefore reducing the validity of any conclusions drawn. In addition, efforts should be made to ascertain whether any symptoms of insomnia are problematic for the child or for the parent, and if it is the latter, whether this is attributed the same clinical relevance.

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CHAPTER TWO

The relationship between sleep and daytime behaviour in children with Autism Spectrum Disorder

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Abstract

Background. Sleep problems have been reported frequently in children with autism spectrum disorder (ASD), with some evidence of an association between sleep problems and daytime challenging behaviour, although the direction of this association is unclear. The aim of this research was to explore the putative temporal association between sleep and daytime presentation.

Methodology. Seventeen children with ASD, aged 5-13 years (mean= 10.1, SD= 2.2), and their families completed questionnaire assessments of both sleep problems and challenging behaviour, as well as participating in a seven-day sleep study. During the sleep study, actigraphy data were collected alongside sleep diaries and twice-daily ratings of challenging behaviour.

Results. Participants who had the highest sleep problem scores scored significantly higher on measures of challenging behaviour. A significant association was observed between night-wakings and overall challenging behaviour score. Linear mixed effects regression analyses identified that time spent awake after sleep onset the previous night, interacted with irritability to significantly predict severity of daytime challenging behaviour during the subsequent day. Sleep parameters did not predict subsequent daytime irritability, and neither irritability nor challenging behaviour predicted sleep the following night.

Discussion. The findings support the existing empirical literature, as well as providing novel evidence for a unidirectional temporal association between poor sleep and subsequent daytime challenging behaviour. Future research should aim to replicate these findings and explore the nature of the relationship between irritability, sleep and behaviour.

2.1 Introduction

Autism Spectrum Disorder (ASD) is a pervasive developmental disorder characterised by impairments in social interaction and communication, and restricted or repetitive behaviours or interests (Diagnostic and Statistical Manual: 5th edition; American Psychiatric Association, 2013). Many individuals with ASD are reported to experience sleep problems, although these difficulties may be overshadowed by other issues perceived as more pressing, such as challenging behaviour (Reynolds & Malow, 2011). Prevalence estimates of sleep disorders in people with ASD vary from 50% to 80% (For a review, see Elrod & Hood, 2015; Richdale & Schreck, 2009) depending on the age and ability of the sample, definition and measurement of the sleep problem, and whether participants have a current or lifetime history of sleep problems. Although there is no definitive explanation for why people with ASD are more likely to experience sleep problems, hypotheses include an altered melatonin response, a genetic basis for a shift in circadian rhythm and atypical levels of arousal (Chamberlain & Herman, 1990; Hu et al., 2009; Melke et al., 2008; Nicholas et al., 2007; Richdale & Prior, 1995; Rossignol & Frye, 2011). Furthermore, some of the other characteristics commonly observed in people with ASD, such as heightened anxiety or arousal, difficulty in interpreting social cues, or preference for routine may also exacerbate sleep problems (Henderson, Barry, Bader & Jordan, 2011; Patzold, Richdale & Tonge, 1998; Richdale & Prior, 1995).

Several types of sleep problem have been observed in people with ASD, including difficulties with sleep onset and maintenance, early waking, and irregular or poor quality sleep, particularly in young children. Other common problems include waking during the night for longer periods of time than is observed in comparison

groups, or having an overall decrease in the total time spent asleep (Honomichl, Goodlin-Jones, Burnham, Gaylor & Anders, 2002; Hoshino, Watanabe, Yashima, Kaneko, & Kumashiro, 1984; Patzold et al., 1998; Richdale & Prior, 1995). These difficulties have been reported as being more prevalent, frequent, and severe than those observed in typically-developing children or other clinical comparison groups, such as individuals with developmental delay (Couturier et al., 2005; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen & Hansen, 2008; Richdale & Prior, 1995; Schreck & Mulik, 2000). A recent meta-analysis by Elrod and Hood (2015) concluded that children with ASD sleep for, on average, 32 minutes less than typically-developing children, take 10.9 minutes longer to fall asleep, and have 1.9% less efficient sleep.

Several reviews have emphasised the importance of identifying not only the topography of the sleep problem but also the possibility of an underlying sleep disorder (Reynolds & Malow, 2011). Cohen, Conduit, Lockley, Rajaratnam and Cornish (2014) posit that the vast range of cognitive abilities and behavioural presentations that are encompassed within the spectrum of ASD is also reflected in the wide variety of sleep problems and disorders. They suggest that the majority of these fall under the classification of insomnia, parasomnia or circadian rhythm sleep-wake disorders, in the International Classification of Sleep Disorders (ICSD-3; American Academy of Sleep Medicine, 2014). Richdale and Schreck (2009) reported that although diagnostic criteria do not always translate well to sleep problems experienced by those with ASD, the most frequently reported difficulties can be best explained by a diagnosis of behavioural insomnia of childhood, in the ICSD-2 (American Academy of Sleep Medicine, 2005). They suggest that this may

be due to younger, or lower-functioning children having developed unhelpful and inflexible bedtime associations, or older and high-functioning children experiencing anxiety, resulting in settling difficulties.

It has been suggested that poor sleep in people with ASD is associated with problematic daytime presentations, such as hyperactivity, social and emotional problems, communication impairments, and increased strength of autistic symptomatology (Allik, Larsson & Smedje, 2006; Hoshino et al., 1984; Malow et al., 2006; Patzold et al., 1998; Schreck, Mulick & Smith, 2004). Furthermore, poor sleep has also been associated with an increase in challenging daytime behaviours, such as aggressive or disruptive behaviour, self-injury and stereotypy (Goldman et al., 2011; Malow et al., 2006; Matson, Ancona & Wilkins, 2008; Schreck et al., 2004). Patzold et al. (1998) found that daytime behaviour problems significantly correlated with night-waking and poor sleep quality, as well as past and current sleep problems. This was replicated by Henderson et al. (2011) who observed that externalising behaviours were negatively correlated with sleep quality, and Tudor, Hoffman and Sweeney (2012), who reported associations between sleep onset delay and sleep duration and stereotypy, as well as between social interaction and the sleep problems of sleep onset delay, sleep duration, night-wakings, parasomnias, and sleep-disordered breathing. Rzepecka, McKenzie, McClure & Murphy (2011) also observed that sleep problems accounted for a large amount of the variance of challenging behaviour, with slightly less accounted for by anxiety. Interventions for sleep problems have demonstrated a positive impact on rates of daytime challenging behaviour in children with and without ASD (Minde, Faucon & Falkner, 1994; Mindell, Kuhn, Lewin, Meltzer & Sadeh, 2006; Wiggs & Stores,

1999). Similarly, Wiggs and Stores (1996) compared daytime behaviour of children with a diagnosis of ASD with and without sleep disturbance, and reported that children with sleep problems were more likely to present with several forms of challenging daytime behaviours, including, stereotypy and hyperactivity, as well as irritability. Heightened irritability or hostility has also been reported as being correlated with sleep duration, night-waking and parasomnia, as well as with sleep anxiety and daytime sleepiness by Mazurek and Sohl (2016).

In combination, these findings imply that a relationship exists between sleep problems and increased daytime challenging behaviour and irritability in people with ASD. However, the majority of existing studies employ a cross-sectional design, and compare behaviour between those above and below a sleep disorder cut-off, or explore associations between measures of sleep and behaviour. Thus, the findings could be interpreted as daytime behaviour and irritability having an impact on sleep, or that an underlying factor exists which influences both of these variables. The lack of longitudinal data is a key limitation of existing literature, as the direction of any temporal association between sleep and daytime presentation cannot be identified. Longitudinal data are necessary in order to design targeted and effective interventions, in that if sleep problems are influencing levels of challenging behaviour and irritability, focusing the intervention on the sleep problems would have positive outcomes for both sleep quality and daytime challenging presentation. By contrast, implementing an intervention for challenging behaviour or irritability alone, would have limited effectiveness in improving sleep, and may not be efficacious in reducing mood or behaviour if the association with sleep problems is particularly strong.

The majority of sleep data for children with ASD are collected via parent- or carer-reports. Whilst parent- or carer-completed measures are an effective method for collecting data from larger samples, and with children who may not be able to tolerate physiological measures, such as polysomnography[1], some research has suggested that parent- and carer-reports may not be accurate. For example, parents may over- or underestimate their child's sleep problems, or be unaware of their child's night-time behaviours, especially if they sleep well themselves (Hering, Epstein, Elroy, Iancu & Zelnik, 1999; Short, Gradisar, Lack, Wright & Chatburn, 2013; Werner, Molinari, Guyer & Jenni, 2008). Actigraphy has been used as an acceptable alternative to polysomnography in the ASD population, as it is an unobtrusive method of estimating the child's sleep behaviour in their home environment. Whilst it is less sensitive than polysomnography, and uses white light and movement as a proxy measure of sleep, it provides a more objective and accurate estimation of a child's sleep than parent-report alone (Sadeh, 2011; Sadeh & Acebo, 2002).

The present study uses a combination of sleep diaries and actigraphy over the course of a seven-night data collection period, in order to generate objective measures of sleep alongside concurrent parental ratings of daytime challenging behaviour and irritability. The aim of the study was to elucidate the temporal relationship between sleep parameters of quality and duration, and daytime challenging behaviour in children with ASD. The first hypothesis was that those reporting greater sleep problems would also present with more challenging

[1] Polysomnography involves the measurement of multiple physiological parameters related to sleep, such as air flow, blood pressure, eye movement, and heart rhythm (Vaughn & Giallanza, 2008).

behaviour. The second hypothesis was that associations would exist between the cross-sectional sleep parameters and ratings of challenging behaviour, as measured using questionnaire data. Finally, it was hypothesised that nights during which children experienced more troubled sleep, as measured using actigraphy, would be associated with more severe daytime ratings for challenging behaviour and irritability. However, it is unclear whether this increase in daytime behaviour and irritability would be observed during the preceding or subsequent day and therefore the direction of the association cannot be predicted.

2.2 Methodology

2.2.1 Participants

Families who had participated in past research projects and who had consented to be contacted for the purposes of future research were sent postal information about the present project. This was then followed up with a telephone call, to determine eligibility and willingness to participate. The project was also advertised at local family conferences dedicated to sleep in individuals with ASD, as well as on the research team's website and social media page. Children aged 4-15 years with any professional diagnosis on the autism spectrum and with any level of intellectual ability were eligible to participate. The child's parents had to consider their child to have a sleep problem to take part, but a formal diagnosis of a sleep disorder was not required.

Twenty families expressed an interest and consented to participate. It was not possible to calculate an accurate participant response rate, as the number of eligible families who had received details of the project was unclear. One family retired from the study partway through data collection, and two children were

excluded due to a high level of missing data. Seventeen children and their families therefore constituted the sample. Children were aged between 5 and 13 years (mean= 10.1, SD= 2.2), and 65% were male. Diagnoses of ASD had been received from paediatricians, local mental health services and Child Development Centres. None of the children had a diagnosis of epilepsy. Five children were currently receiving medication for sleep problems, one child was taking a psychostimulant and two children were taking antihistamines. All children lived at home with their biological mother, and either their biological father or step-father. Participant characteristics are summarised in Table 2.1 and Appendix 6.

Table 2.1 – Participant characteristics

Participant	Age (Years)	Gender	Performance IQ[1]	Adaptive Behaviour[2]
1	9	Male	NA	56
2	12	Female	112	68
3	7	Female	95	89
4	8	Male	131	75
5	13	Male	89	76
6	13	Male	94	79
7	11	Female	111	78
8	8	Male	110	80
9	12	Male	110	74
10	10	Male	112	62
11	9	Male	70	68
12	10	Male	76	66
13	7	Female	77	126
14	11	Female	112	68
15	10	Male	127	97
16	5	Male	>103	65
17	10	Female	84	67

[1] As measured on the Wechsler Abbreviated Scale of Intelligence
[2] As measured on the Vineland Adaptive Behavior Scales

Descriptive statistics for adaptive functioning as measured on the Vineland Adaptive Behavior Scale (VABS), are given in Table 2.2. The majority of children fell within the 'low' or 'low to moderate' ranges across all subscales, except for motor skills in which most children scored within the 'adequate' range. Of the seventeen participants, eleven met the threshold for autism on the Autism Diagnostic Observation Schedule (ADOS), and the remaining six met criteria for autism spectrum disorder. One child was classed as having an intellectual disability based on performance on the Mullen Scales for Early Learning (MSEL; Age equivalent scores 23-36 months). Another child scored at ceiling on the MSEL, yet was below the recommended age range on the Wechsler Abbreviated Scales of Intelligence (WASI), and so standardised IQ data were not available. Estimation of intellectual ability for the remainder of the sample was based on performance IQ due to a significant discrepancy observed between verbal and performance subscales for a number of participants, and the likelihood of overall IQ being underestimated as a result (Happé, 1994; Siegel, Minshew & Goldstein, 1996). The performance IQ mean for the remainder of the sample was therefore 100.6 (SD= 18.6).

Table 2.2 - Mean scores for each subscale of the Vineland Adaptive Behavior Scales and overall adaptive behaviour score, and percentage of sample falling within each category denoting level of functioning.

	N	Mean *(SD)*	% (n)				
			Low	Low - moderate	Adequate	High - moderate	High
Communication	17	85.1 *(16.9)*	23.5 (4)	35.3 (6)	29.4 (5)	11.7 (2)	0 (0)
Daily Living Skills	17	78.5 *(22.1)*	41.2 (7)	35.3 (6)	17.6 (3)	0 (0)	5.9 (1)
Socialisation	17	71.7 *(17.5)*	58.8 (10)	29.4 (5)	5.9 (1)	5.9 (1)	0 (0)
Motor Skills	17	89.6 *(20.2)*	23.5 (4)	23.5 (4)	47.1 (8)	5.9 (1)	0 (0)
Overall Adaptive Behaviour	17	76.1 *(16.3)*	47.1 (8)	41.2 (7)	5.9 (1)	5.9 (1)	0 (0)

2.2.2 Measures

Measures were completed either over the telephone, in person with the child or parent, or as part of a questionnaire pack.

Vineland Adaptive Behavior Scales (VABS: Sparrow, Balla & Cicchetti, 1984). The VABS was used to assess the child's functional ability across domains of communication, daily living, socialisation and motor skills, and provides a standardised score for overall adaptive behaviour. This was completed two weeks prior to the family attending the university. The VABS has been demonstrated as having sufficient internal consistency, test-retest reliability and inter-rater reliability with typically-developing samples as well as those with developmental delay (De Bildt, Kraijer, Sytema & Minderaa, 2005; Sparrow et al., 1984).

Modified Simonds and Parraga Sleep Questionnaire (MSPSQ; Simonds & Parraga, 1982; Wiggs & Stores, 2004. Appendix 2). The original measure, published in 1982, is completed by the parent, and is used to assess sleep quality and identify sleep disturbances in children. The modified version has been adapted for use with children with developmental disorders, and can be used to derive subscale scores for bedtime resistance, sleep onset delay, sleep anxiety, night-waking, parasomnia, sleep-disordered breathing and daytime sleepiness, as well as an overall score. The scoring methods suggested by Johnson, Turner, Foldes, Malow and Wiggs (2012), and Wiggs and Stores (2004) have been used.

Autism Diagnostic Observation Schedule (ADOS: Lord, Rutter, DiLavore & Risi, 2001). The ADOS is a commonly-used standardised observational instrument, which makes use of activities designed to engage the individual in social interaction, in order to assess for common features of ASD. The

ADOS has good inter-rater reliability and predictive validity, and specificity and sensitivity rates above 87% (Lord et al., 2001). The measure was included to confirm the child's existing diagnosis of ASD.

Mullen Scales of Early Learning (MSEL; Mullen, 1995). The MSEL is a measure of cognitive ability for children aged 0-68 months, and is also commonly used with individuals with developmental disabilities. The measure includes subscales assessing gross and fine motor skills, expressive and receptive language ability and visual reception, and has been reported to have adequate reliability and validity (Bishop, Guthrie, Coffing & Lord, 2011)

Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999). The WASI is a brief measure of intellectual functioning designed for typically-developing individuals aged 6-89.9 years, consisting of four subscales; vocabulary, similarities, block design and matrix reasoning. The WASI was only administered if the child scored at ceiling on the MSEL. Psychometrics derived from the standardisation sample demonstrated correlation coefficients above .84 when compared to the full scale measure (Wechsler, 1999).

Amended Challenging Behaviour Questionnaire (Amended CBQ. Appendix 3). Parents were asked to complete a measure which had been modified to combine the Challenging Behaviour Interview (CBI; Oliver et al., 2003) and the Challenging Behaviour Questionnaire (CBQ; Hyman, Oliver & Hall, 2002), referred to here as the Amended CBQ. Both measures have demonstrated adequate reliability and validity individually (Hyman et al., 2002; Oliver et al., 2003). This measure identified the presence of challenging behaviours across domains of self-injury, aggression, destruction of property and stereotypy, as well as other

behaviours. The behaviours within each domain were then given a subscale rating based on their pervasiveness, i.e. the duration and frequency of the behaviour, and how often the behaviour required intervention from the parent.

Questionnaire about Behavioural Function (QABF; Paclawskyj, Matson, Rush, Smalls & Vollmer, 2000; Richards, 2015). The domain that received the highest subscale score on the amended CBQ was then the focus of the QABF. This questionnaire presents multiple scenarios and asks the parent to rate the likelihood of their child presenting with the chosen behaviour in each situation. From this, possible functions of the behaviour are identified. The QABF has been demonstrated as a valid alternative to experimental functional analysis, and has reportedly strong psychometric properties (Matson, Tureck & Rieske, 2012). The version presented here, has been amended to include additional subscales focused on ASD-specific sensory-based and routine-based functions (Richards, 2015).

Actigraphy. Families agreed to a week during which they were available to complete the main sleep assessment. During this week, the child's sleep was measured using sleep diaries completed by the parent (Appendix 4), and an actiwatch that was worn on the child's non-dominant wrist (Philips Respironics, 2014). Actigraphy is a method used to estimate sleep duration and quality, based on the wearer's movement and the presence of white light. The software uses this information to calculate sleep intervals, and generate statistics such as sleep efficiency, total sleep time, and sleep onset latency. Actigraphy has been used in several studies with children with ASD, and has shown acceptable consistency with parent-completed sleep diaries (Allik et al., 2006; Goodlin-Jones, Sitnick, Tang, Liu

& Anders, 2008; Wiggs & Stores, 2004). Here, the automatically generated data were checked against sleep diaries completed by parents, and cleaned according to an agreed protocol (Appendix 5).

An additional sleep parameter was calculated based on the parent-reported bedtime, and the time at which the actigraphy estimated the child to have fallen asleep. This was due to the sleep onset latency parameter being defined by the actigraphy, as the time during which the individual is trying to fall asleep. This normally corresponds to the latency between the individual turning off the lights and them falling asleep, and is represented by a period of rest on the actigraphy output. However, it was considered that in our sample, the time at which the child goes to bed is determined primarily by the parent. It may be that, despite being in bed, the child is not necessarily trying to sleep at this time and is therefore active for a period before settling down to sleep, the latter of which would be detected by the actigraphy as sleep onset latency. However, the active time between bedtime and settling time, is also likely to be an important feature of sleep in children with ASD, and therefore this additional parameter of the latency between parent-intended bedtime and time falling asleep was included.

In addition to measuring the child's sleep, the sleep diaries included items asking parents to rate the severity of their child's daytime irritability and challenging behaviour in the morning and afternoon, as well as listing any naps that the child took during the day. Once the data had been collected, the family was provided with a report which summarised their child's sleep data, and their performance on the other measures.

2.2.3 Analysis

Questionnaire packs were returned for fifteen of the seventeen participants. Data for the direct measures were available for the whole sample. Analyses were conducted using the Statistical Package for the Social Sciences: Version 19 (SPSS: IBM Corp., 2010), R (R Core Team, 2015), and the lme4 package (Bates, Mächler, Bolker & Walker, 2014). Where data were non-normally distributed, non-parametric statistical methods were used. An alpha level of p=.01 was employed to account for multiple comparisons.

2.3 Results

2.3.1 Challenging Behaviour

All of the children displayed between one and seventeen forms of behaviour which parents considered to be challenging (mean= 5.6, SD= 4.3). Scores from the amended CBQ are summarised in Table 2.3. Stereotyped behaviour was the most prevalent topography, with 76.5% of children presenting with some form of stereotyped behaviour.

Table 2.3 - Mean score for each topography of challenging behaviour in the amended CBQ, and the proportion of the sample presenting with each behaviour (Summation of n may exceed N, as participants present with several behaviours).

	N	% of sample (n)	Mean *(SD)*
SIB	17	52.9 (9)	3.8 *(4.3)*
Hitting self		23.5 (4)	
Hitting self with object		11.8 (2)	
Biting self		23.5 (4)	
Pulling at hair/ skin		5.9 (1)	
Scratching self		11.8 (2)	
Inserting objects		5.9 (1)	
Other		5.9 (1)	
Aggressive Behaviour	17	58.8 (10)	4.1 *(4.2)*
Hitting		41.2 (7)	
Hitting with object		11.8 (2)	
Pulling or grabbing		23.5 (4)	
Scratching or pinching		5.9 (1)	
Spitting		5.9 (1)	
Verbally aggressive		41.2 (7)	
Other		5.9 (1)	
Destructive Behaviour	17	47.1 (8)	3.4 *(4.1)*
Tearing or ripping items		17.6 (3)	
Throwing, kicking, smashing small items		17.6 (3)	
Slamming or kicking doors		41.2 (7)	
Tipping, throwing, smashing large items		17.6 (3)	
Pulls items from shelves or walls		5.9 (1)	
Other		5.9 (1)	
Stereotypy	17	76.5 (13)	5.5 *(4.0)*
Repetitive whole body movement		23.5 (4)	
Repetitive movement of an object		23.5 (4)	
Repetitive movement of body part		41.2 (7)	
Visual regard e.g. strobing		17.6 (3)	
Mouthing		35.3 (6)	
Other e.g. clearing throat, grunting, jumping		23.5 (4)	

Other	17	47 (8)	3.0 *(3.9)*
Pica		5.9 (1)	
Vocalisations		29.4 (5)	
Smearing		5.9 (1)	
Stealing		17.6 (3)	
Other		11.8 (2)	

Sixteen of the participants had complete data for the QABF. These data demonstrated that the behaviours identified in the amended CBQ were maintained by numerous functions, and that some participants' behaviour was maintained by more than one function. Mean scores for each of the subscales are presented in Table 2.4.

Table 2.4 - Mean scores for each subscale of the QABF.

Function of behaviour	N	Mean (SD)
Attention	16	5.5 (5.1)
Task Escape	16	7.6 (5.2)
Self-stimulation	16	9.3 (3.6)
Pain or Discomfort	16	5.6 (4.1)
Tangibles	16	6.9 (5.1)
Sensory	16	9.1 (4.3)
Routine	16	9.7 (3.8)
Social Escape	16	8.3 (4.3)

2.3.2 Sleep Problems – Questionnaire Data

Descriptive statistics for the MSPSQ data demonstrated that 87% of the sample scored above the recommended sleep disorder cut-off of 56 (Johnson et al., 2012), suggesting that the majority of the sample met criteria for a sleep disorder worthy of clinical intervention. Analysis of the descriptive statistics of the subscale scores, where any individual item scoring four or above is suggestive of the frequent occurrence of a particular sleep problem (Wiggs & Stores, 2004), demonstrated that

the most prevalent sleep problems fell under the category of bedtime resistance. When applying the threshold of four to the median scores for each subscale, daytime sleepiness was observed to score most highly. The summary statistics for the MSPSQ are given in Table 2.5.

Table 2.5 - Summary statistics for the MSPSQ.

	N	Mean *(SD)*	% over median cut-off (n)	% over single item cut-off (n)
MSPSQ	15	73.3 *(14.6)*		
Bedtime Resistance		2.6 *(0.9)*	26.7 (4)	86.7 (13)
Sleep Onset Delay		3.2 *(1.1)*	40.0 (6)	40.0 (6)
Sleep Anxiety		2.3 *(0.7)*	6.7 (1)	80.0 (12)
Night-waking		2.7 *(0.9)*	6.7 (1)	53.3 (8)
Parasomnia		1.8 *(0.5)*	0 (0)	80.0 (12)
Sleep-disordered Breathing		1.6 *(0.9)*	6.7 (1)	40.0 (6)
Daytime Sleepiness		3.0 *(1.3)*	33.3 (5)	66.7 (10)

No significant Spearman's correlations were observed between the sleep variables, as measured on the MSPSQ, and adaptive functioning, as measured on the VABS. To investigate the association between sleep and challenging behaviour, a median split was carried out based on the MSPSQ total score. A significant difference was observed in the total scores of the amended CBQ when a t-test was conducted between the group with high MSPSQ scores (mean= 24.0, SD= 13.7) and those with low MSPSQ scores (mean= 11.7, SD= 5.4) ($t(9.4)= -2.34$, $p= .04$).

Spearman's correlations were then conducted between the MSPSQ sleep disorder subscales and the function subscales of the QABF. A significant correlation was observed between daytime sleepiness and the self-stimulatory scale ($r= .70$, $p< .01$).

Considering the Spearman's correlations between the sleep disorder subscales on the MSPSQ and the challenging behaviour scores on the amended CBQ, night-wakings were observed to show a significant positive association with the overall score for the amended CBQ (r= .62, p= .01).

2.3.3 Sleep Problems – Actigraphy Data

Each child had valid actigraphy data for between four and eight nights. The sleep parameters included the times at which it was estimated that the child went to bed at night and got out of bed in the morning, the total time that the child spent in bed and the time spent asleep, the latency between getting into bed and falling asleep, the amount of time spent awake after sleep onset (WASO), and the number of wakings during the night. The additional parameter of the latency between parent-reported bedtime and falling asleep was also analysed. The actiware also provides an estimate of the child's sleep efficiency as a percentage, based on a combination of the above variables. Summary statistics for the actigraphy data are presented in Table 2.6.

Table 2.6 - Summary statistics for the sleep parameters, as measured using actigraphy.

	N	Mean *(SD)*	Minimum	Maximum
Bedtime (hh:mm:ss)	17	21:17:11 *(01:06:30)*	19:34:25	23:45:35
Get-Up time (hh:mm:ss)	17	06:40:24 *(00:52:46)*	05:07:00	08:35:05
Time in bed (hh:mm:ss)	17	09:23:12 *(00:40:26)*	08:26:21	10:41:30
Time asleep (hh:mm:ss)	17	08:09:11 *(00:34:27)*	06:57:47	09:13:12
Bedtime-sleep latency (hh:mm:ss)	15	00:50:33 *(00:35:24)*	00:07:26	01:49:21
Sleep onset latency (mins)	17	9.51 *(8.56)*	0.71	35.33
WASO (mins)	17	50.48 *(16.26)*	25.50	79.07
Frequency of awakenings per night	17	38.81 *(9.75)*	25.57	64.50
Sleep Efficiency (%)	17	86.93 *(2.59)*	82.35	91.40

As with the MSPSQ data, investigation of the Spearman's correlations between sleep parameters, as measured using actigraphy, and adaptive functioning, as measured on the VABS, identified no significant associations. Spearman's correlations were then carried out between the actigraphy parameters and the challenging behaviour subscales on the amended CBQ, with no significant associations identified.

2.3.4 Temporal Associations between Sleep and Challenging Behaviour

Linear mixed effects analyses were conducted to study the temporal relationships between the actigraphy sleep parameters and the daytime challenging behaviour and irritability data, as rated by parents in the sleep diaries. Estimates and standard errors for each parameter were calculated using maximum likelihood estimators (M estimators). This method of analysis is more robust than sum of squares indicators, as they are less reliant on the assumptions of parametric statistics. Furthermore, the method allows for the clustering of data across multiple levels, and in this manner accounts for autocorrelation occurring in longitudinal repeated measures designs. In the present study, challenging behaviour data were collected in both the morning and afternoon, for each day of the week, and for each participant. This resulted in three levels of measurement: time of day, day of week, and participant number, and therefore several potential sources of autocorrelation. The analysis accounted for differences at the level of intercept, to allow for differences in the sleep parameters between participants (Pinheiro & Bates, 2006). The intercept model was chosen, rather than slope, due to the sample size. This was because the number of terms required to model differences in slope would have greatly exceeded the number of data-points.

Does poor sleep predict daytime irritability? First, the extent to which the levels of the longitudinal study design and the individual differences between participants contributed to the variance in severity ratings of daytime irritability was analysed. To do this, variables coding for participant number, whether each time-point fell on a weekday or weekend, and whether data were collected in the morning or afternoon were each added into the null hypothesis model and removed in turn, to determine whether they should be included as random effects in the final model. A significant random effect was identified for the participant variable (X^2= 162.6, p< .001), but not for day (X^2= 0.0, p= 1.0), or time of day (X^2= 0.4, p= 0.5). As a result, the variable denoting participant was included as a random effect in the null hypothesis model.

The fixed effects of total sleep time, WASO and sleep efficiency were then added to the random effects model consisting of the participant variable, and this was compared to the null hypothesis model using a likelihood ratio test. None of the predictors offered significant improvement to the null hypothesis model (X^2= 3.7, p= 0.3), nor were the individual parameter estimates significant (Table 2.7), and so no fixed effects were added to the final model[1]. This implies that variance in daytime irritability is best explained by individual differences between participants, and was not further influenced by sleep time or quality.

Table 2.7 - Fixed effects for the linear mixed effects model predicting irritability.

	Estimate	Standard Error	t
Intercept	-0.86	1.83	-0.47
Total Sleep Time	-0.00	0.00	-0.62
WASO	0.01	0.00	1.31
Sleep Efficiency	0.04	0.02	1.89

[1] Model = **lmer**(Irritability ~ 1 + (1|Participant Number), REML=FALSE, data=d)

Does poor sleep predict daytime challenging behaviour? As with the previous analysis, participant number, day and time of day were entered as random intercepts into the null hypothesis model, but only participant number was retained (X^2= 104.2, p< 0.001), with the random effects of day (X^2= 0.0, p= 1.0) and time of day (X^2= 0.0, p= 1.0) being removed.

The full model including predictor variables of total sleep time, WASO, sleep efficiency and irritability, was then compared to the null hypothesis model, in order to evaluate the fixed effects. Both the removal of WASO (X^2= 6.4, p= 0.01) and irritability (X^2= 62.6, p< 0.001) resulted in a significant change to the model, but total sleep time (X^2= 2.1, p= 0.15) and sleep efficiency (X^2= 3.2, p= 0.07) did not. WASO and irritability were therefore included as fixed effects in the final model[1]. This produced a final model for challenging behaviour, including participant number as a random effect, and WASO and irritability as fixed effects (Figures 2.1 and 2.2). This implies that daytime challenging behaviour is best predicted by WASO and irritability, when accounting for individual difference between participants. Covariance between WASO and irritability was r=-.02, suggesting that there was negligible shared variance between the variables.

Table 2.8 - Fixed effects for the linear mixed effects model predicting challenging behaviour.

	Estimate	Standard Error	t
Intercept	0.26	0.26	1.02
Irritability	0.48	0.05	8.75
WASO	0.01	0.00	1.71

[1] Model = **lmer**(Challenging behaviour ~ 1 + WASO + Irritability + (1|Participant Number), REML=FALSE, data=d)

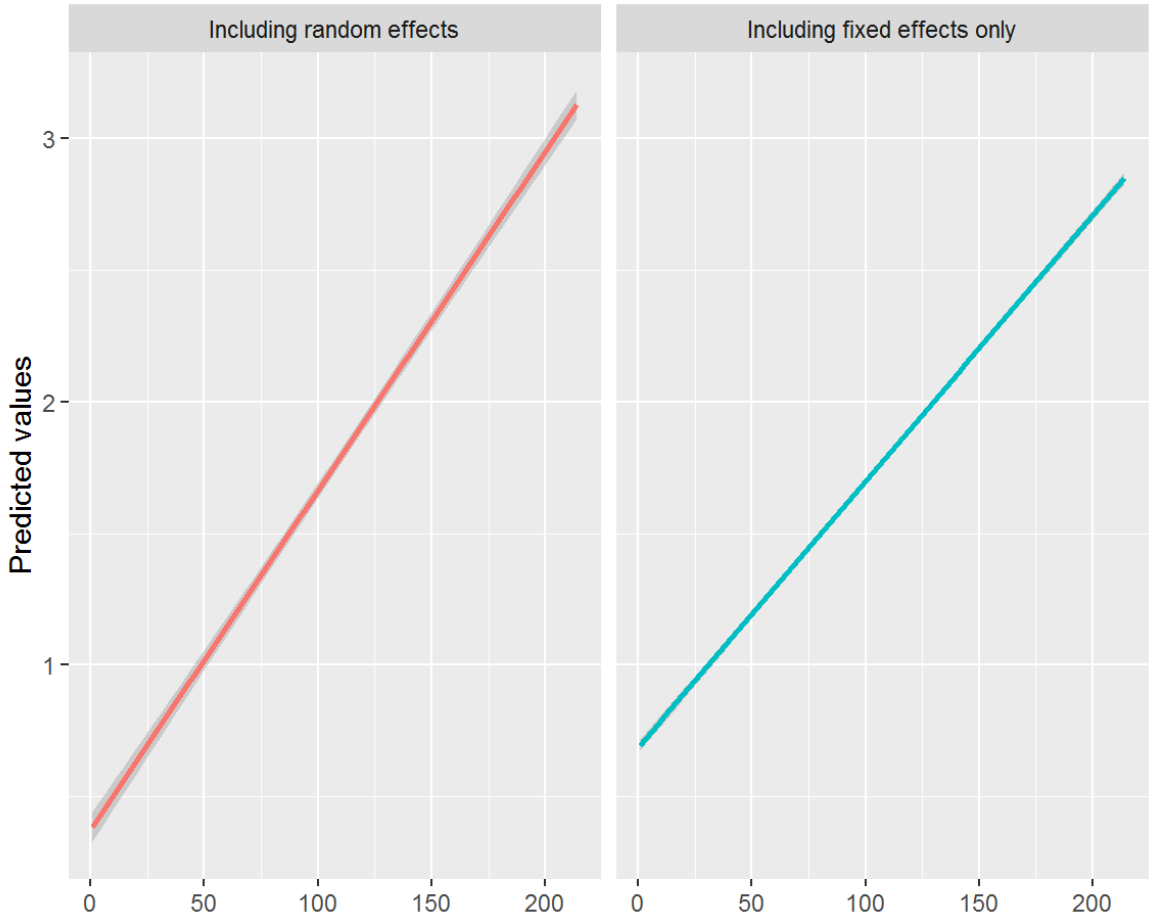

Figure 2.1 - Predicted values for the linear mixed effects model predicting severity of challenging behaviour, conditioned on random and fixed effects only.

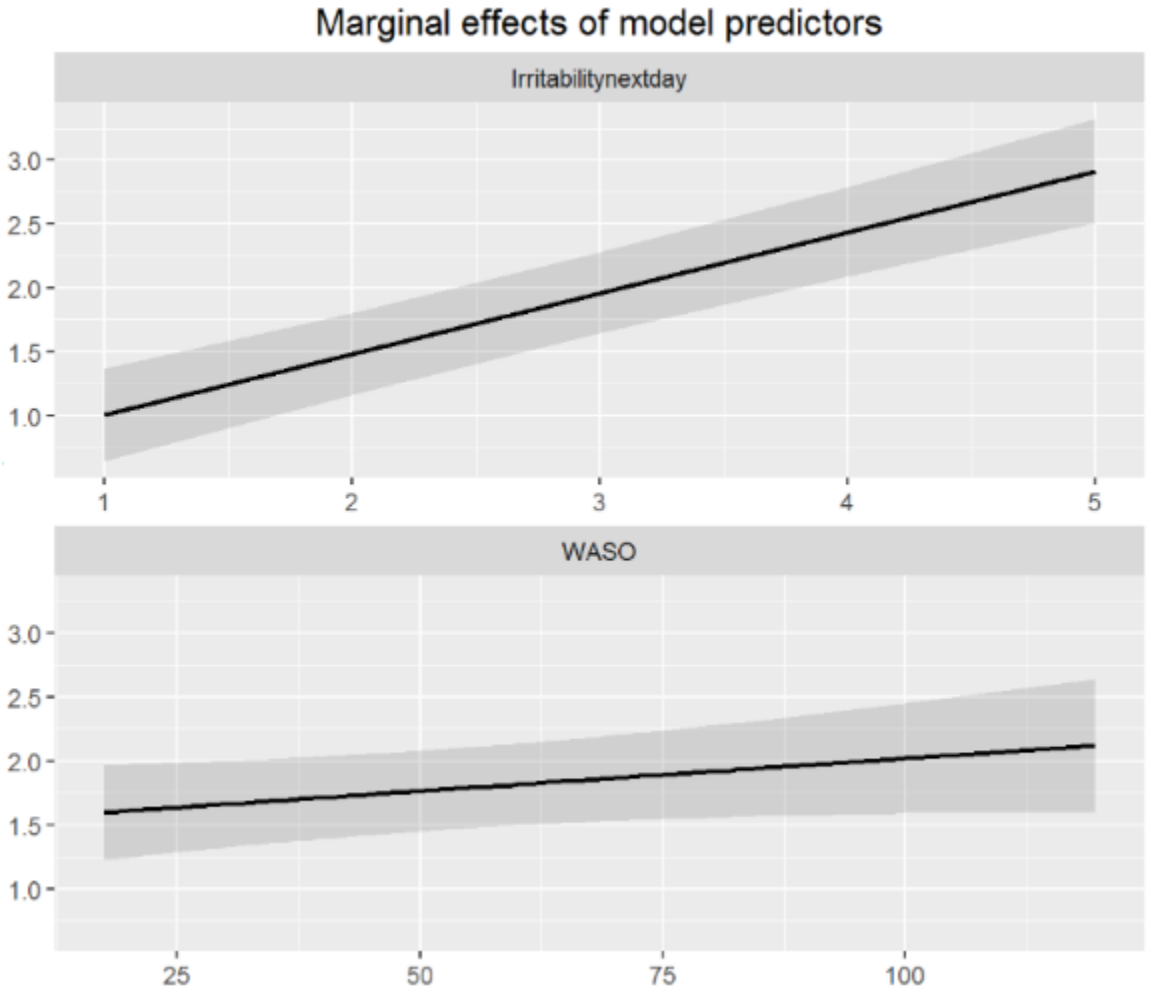

Figure 2.2 – Marginal effects for the predictors of irritability and WASO on ratings of challenging behaviour.

Does daytime irritability or challenging behaviour predict sleep efficiency? Random effects of participant and whether the day was a weekday or weekend were entered into the null hypothesis model. Time of day at which the data were collected was not entered in this analysis, as only data collected in the afternoon were thought to influence sleep efficiency that night, and therefore only afternoon data were included in the analysis. As in the previous analyses, a significant random effect was not observed for day (X^2= 0.0, p= 1.00), and so only

participant number was retained as a random effect in the null hypothesis model (X^2= 8.2, p< 0.01).

Challenging behaviour and irritability ratings were added as fixed effects in the full model, and compared to the null hypothesis model consisting of the participant number. The addition of the fixed effects added no significant contribution to the null model (X^2= 3.3, p= 0.19), and demonstrated no significant individual parameter estimates (Table 2.9). Therefore, no fixed effects were included to the final model[1]. This suggests that variance in sleep efficiency is best explained by individual differences between participants, and was not further influenced by daytime challenging behaviour or irritability.

Table 2.9 - Fixed effects for the linear mixed effects model predicting sleep efficiency.

	Estimate	Standard Error	t
Intercept	88.76	1.24	71.81
Challenging Behaviour	-0.70	0.57	-1.23
Irritability	-0.16	0.49	-0.33

2.4 Discussion

The aim of the present study was to delineate the nature of the relationship between sleep and daytime challenging behaviour in individuals with ASD. The study sought to improve upon previous work by including a temporal analysis of sleep and daytime challenging behaviour. The results support the hypothesis that poor sleep quality has a negative impact on ratings of daytime behaviour the following day. Specifically, the time spent awake after having fallen asleep (WASO), in interaction with parent-reported irritability, are significant predictors of the severity of daytime

[1] Model = **lmer**(Sleep Efficiency ~ 1 + (1|Participant Number), REML=FALSE, data=d)

challenging behaviour. Although numerous authors have hypothesised that observed associations between sleep problems and daytime challenging behaviour in cross-sectional datasets are indicative of a negative outcome of poor sleep on daytime functioning, this study provides the first findings based on longitudinal data to support this argument. Furthermore, the observation that daytime behaviour does not explain variance in sleep quality the subsequent night, further implies that the association between these two variables is unidirectional.

Notably, it was specifically the time spent awake during the night, as measured using actigraphy, that was predictive of subsequent challenging behaviour, in contrast to other predictors, such as sleep duration. This suggests that it is disturbed or broken sleep rather than shortened sleep duration or sleep efficiency which is causally related to challenging behaviour for children with ASD. This assertion is supported by the correlations between sleep variables as measured by the MSPSQ, between reported night-wakings, and both overall score on the amended CBQ, and total frequency of challenging behaviours.

Other observed associations, although not statistically significant, included the non-social operant functions for challenging behaviour of self-stimulation and pain, and scores for parasomnia. This implies that children whose behaviours are maintained by the non-social functions of self-stimulation or discomfort, also are more likely to show signs of parasomnia. It could be that an underlying factor mediates the relationship between these variables, such as the presence of health problems. The discomfort caused by a health problem may exacerbate behaviours maintained by underlying functions of pain and self-stimulation, in order to reduce or tolerate the distress. However, the discomfort may also increase the likelihood of

the child presenting with signs of parasomnia, such as bruxism or restlessness, and so, in this manner, an additional underlying factor may be influencing scores on both the QABF and MSPSQ. As Goldman et al. (2011) discuss, the parasomnia subscale consists of a wide range of different problems, which can frequently occur independently. In this sense, analysing parasomnias as a subscale of related sleep problems may not yield the most meaningful data, as a child may not score particularly high on the subscale average, yet may still present with behaviours significant enough to reach the threshold recommended by the ICSD. It can be observed that 80% of the sample frequently experienced at least one form of parasomnia, and yet this may not have been reflected in the use of the median data when correlating sleep subscales against challenging behaviour topography and function. Another observed association was between daytime sleepiness and behaviour maintained by a self-stimulatory function, suggesting that children who are tired in the daytime may also present with stereotyped behaviours, such as rocking or hand-flapping. This could imply that the stereotypy acts as a self-soothing behaviour to reduce the distress experienced when feeling tired, or the findings could also be related to the observation that some children become more overactive when tired, rather than more lethargic. It is not possible to test these hypotheses within the constraints of the data presented here, but is a possible avenue for future research.

The potential existence of associations between specific sleep problems and the topographies of challenging behaviour partially support the findings of Tudor and colleagues (2012), who carried out similar correlational analyses. The authors suggested that specific sleep problems may show different relationships with certain

forms of daytime impairment, as they observed relationships between sleep onset delay and sleep duration and stereotypy, whereas social interaction was also related to night-wakings, parasomnias, and sleep-disordered breathing. Similarly, Patzold and colleagues (1998) found that difficult daytime behaviour was correlated with sleeping in the same bed as parents, past or present sleep problems, poor sleep quality and night-wakings, and that shorter sleep duration predicted severity of autistic symptomatology. Furthermore, Mazurek and Sohl (2016) observed multiple significant correlations between sleep disorders and challenging behaviours of aggression, hostility/ irritability, inattention and hyperactivity. However, they observed that sleep anxiety was the best predictor of hostility/ irritability, whereas night-wakings had the strongest association with the remaining three of the four behaviour variables. It would appear that quality of sleep, influenced by features such as night-wakings, restlessness or parasomnias, has a different impact on children's daytime presentation in comparison to reduced sleep time, characterised by longer sleep latencies or early morning awakening. This implies that the nature of the relationship between sleep and daytime presentation may differ between specific sleep problems.

As Wiggs and Stores (2004) identify, a number of research studies identify sleep problems in children with ASD, but do not link these data with underlying diagnostic sleep disorder classifications, which would better inform intervention for these children. As such, a sleep problem could be considered a manifestation of a number of underlying disorders, each of which would be best addressed by different approaches to treatment, and may therefore have different implications on wider functioning. For example, night-waking underpinned by anxiety, may have a

different relationship to daytime behaviour than night-waking caused by an underlying sleep apnoea. Furthermore, the intervention for each of these sleep disorders would differ considerably. Although the present study alluded to the presence of sleep disorders within the sample, a diagnostic measure was not used, and the sleep disorder data taken from the subscales of the MSPSQ should therefore be interpreted with caution. However, it would be an interesting development to apply more stringent diagnostic criteria to the sleep problems observed in a sample of children with ASD, and to replicate the analysis conducted here, to identify whether the observed relationships between sleep disorders and the topographies and functions of challenging behaviour remain.

In addition to advancing our understanding of the nature of the relationship between poor sleep and daytime challenging behaviour, the present study also replicated findings from a number of previous studies, in that children who were reported to have the most significant sleep problems, also scored highest on measures of challenging behaviour (Malow et al., 2006; Wiggs & Stores, 1996). Furthermore, sleep problems were not observed to be related to levels of adaptive functioning, which corresponds to the findings of authors, such as Patzold et al. (1998) who observed that children with ASD experienced more sleep problems then a typically-developing control group, despite being matched for age and IQ, Krakowiak and colleagues (2008) who reported that adaptive functioning was predictive of neither quality nor duration of sleep in their ASD sample, and Mayes and Calhoun (2009) who observed no relationship between intellectual or neuropsychological functioning and sleep disturbance. This would suggest that adaptive or intellectual ability levels are not related to sleep problems in this group,

yet, research in this area appears to be inconclusive, as other authors have reported findings to the contrary (Giannotti et al., 2008; Taylor, Schreck & Mulick, 2012).

Despite there being a number of interesting findings in the present study, a key limitation was the wide range of individual differences within the group, resulting in a relatively non-homogenous sample. Because different types of underlying sleep disorder and topographies of challenging behaviour may be more prevalent in various subgroups within the sample, such as those with and without intellectual disability, or those with autism compared to ASD, it may be that different relationships exist between these variables for different children within the sample. As Cohen and colleagues (2014) discuss, the presentation of children with ASD differs significantly between those who are considered high-functioning and those who have an intellectual disability. Due to the size of our sample, and the difficulty with assessing IQ in a manner that was appropriate for both high- and low-functioning individuals, these two subgroups could not be analysed separately. Similarly, the sample consisted of children displaying a wide range of autistic symptomatology, and with diagnoses of autism and autism spectrum conditions. This limits the extent to which the findings can be generalised beyond this sample, as the association between autistic symptomatology and sleep problems cannot be ascertained and therefore accounted for. However, it should be noted that by analysing the data using a mixed effects linear regression, and including participant in the random effects element of the model, this will have largely accounted for the variance explained by individual differences between the participants. Future research should recruit a large enough sample, such that subgroups within the sample can be analysed separately and compared, such as ASD compared to

autism, or those with and without intellectual disability. Had a larger sample been obtained, it may have been possible to better disentangle the features of ASD that might mediate the relationship between sleep problems and challenging behaviour.

Another limitation of the study is that the findings only provide information about the association between sleep and daytime challenging behaviour, in children whose parents consider them to have a sleep problem. This association may be different for children who display challenging behaviour but do not present with sleep problems. Future studies could assess sleep quality and duration in children with and without sleep problems, and examine temporal associations with daytime challenging behaviour, or other variables, such as attention, affect, or sociability.

In summary, the findings presented here contribute to an improved understanding of the association between sleep and daytime challenging behaviour, in that the time spent awake after sleep onset, in combination with irritability, predicted the severity of challenging behaviour the following day. Although we cannot conclude that sleep problems cause daytime challenging behaviour, it is likely that disturbed sleep may exacerbate behaviour, and that by helping to improve a child's sleep, we may also observe positive changes in their daytime behaviour. This finding therefore has implications for the way in which clinicians assess for the contributors to challenging behaviour, and the way in which interventions are developed. Future research should aim to replicate these findings with a larger, more homogenous sample.

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CHAPTER THREE

Public Dissemination Document

3.1 Literature Review

3.1.1 Background

Insomnia is a sleep disorder characterised by difficulty initiating sleep (DIS), difficulty maintaining sleep (DMS) and early morning awakening (EMA), with evidence of associated distress or daytime impairment (Diagnostic and Statistical Manual: Fifth edition, American Psychiatric Association, 2013; International Classification for Sleep Disorders: Third edition; American Academy of Sleep Medicine, 2014). A wide range of prevalence estimates for insomnia in the child and adolescent population have been reported, from 4% to 41% (Archbold, Pituch, Panabi & Chervin, 2002; Camhi, Morgan, Pernisco & Quan, 1999). This varies dependent on the way in which insomnia has been defined, the methodology employed to identify the presence of insomnia, and the population from which the research sample was drawn. Consequently, it is difficult to gauge the extent to which insomnia is of clinical importance in this age-group. Furthermore, it has been questioned whether the diagnostic criteria can been meaningfully applied to this population, and whether insomnia should be considered in a more descriptive context with children and adolescents, rather than as a diagnostic term (Glaze, Rosen & Owens, 2002; Meltzer & Mindell, 2014; Mindell et al., 2006).

Insomnia has been identified as being more common in physical and mental health disorders, yet the lack of synthesised prevalence data in the non-clinical population limits the extent to which the prevalence of insomnia in clinical groups can be accurately compared. This means that it is difficult to estimate the extent to

which clinical populations are at an increased risk of presenting with insomnia. The range of definitions and methods of assessment in the empirical literature also limits the extent to which the findings can be used to understand the components of insomnia, and therefore its value in developing effective interventions. The aims of the study were therefore to provide an overall estimate for the prevalence of insomnia in the non-clinical population of children and adolescents, adjusted for the quality of the research methodology. The study also looked to compare the prevalence figures identified for the non-clinical population, with those identified for populations of children and adolescents with physical and mental health conditions.

3.1.2 Methodology

A search of the research literature was conducted to identify articles that reported the prevalence of insomnia in children and adolescents aged 4-18 years. These articles were screened against a number of eligibility criteria (Table 3.1). Articles that did not meet these criteria were excluded. Additional criteria were developed to construct a quality framework, based on Munn, Moola, Lisy and Riitano (2014). This was used to test the quality of each article, with articles considered to be of better methodological quality receiving a higher weighting. The prevalence data from each article were then pooled to generate overall prevalence estimates, using a random-effects model, and then a quality-effects model based on the quality weightings. This was repeated for each definition of insomnia, and for different clinical populations.

Table 3.1 - Strategy for screening articles.

Criteria	Articles to be excluded
English language	Non-English language
Empirical study	Reviews, guides, conference proceedings, case studies etc.
Typically-developing sample	Sample includes children with a neurodevelopmental disorder
Sample of school-age	Sample includes adult or infant populations
Relevant	Irrelevant articles, such as pharmacological trials
Non-biased recruitment	Sample recruited on basis of having a sleep, or related, disorder
Prevalence statistics	Frequency or prevalence data cannot be extracted

3.1.3 Findings

The findings are summarised in Figure 3.1. The pooled meta-analyses identified a prevalence estimate of 16-20% for the presence of insomnia in the child and adolescent population. The prevalence estimates for each of the three symptoms were 14% for DIS, 9% for DMS and 6% for EMA. More stringent variations of the insomnia criteria, such as the inclusion of distress or daytime impairment, or the exclusion of comorbid disorders, reduced the pooled prevalence estimates to 10% and 4% respectively. Fifty-four to 61% of children and adolescents who were considered to have anxiety disorders, depression or chronic pain also presented with insomnia.

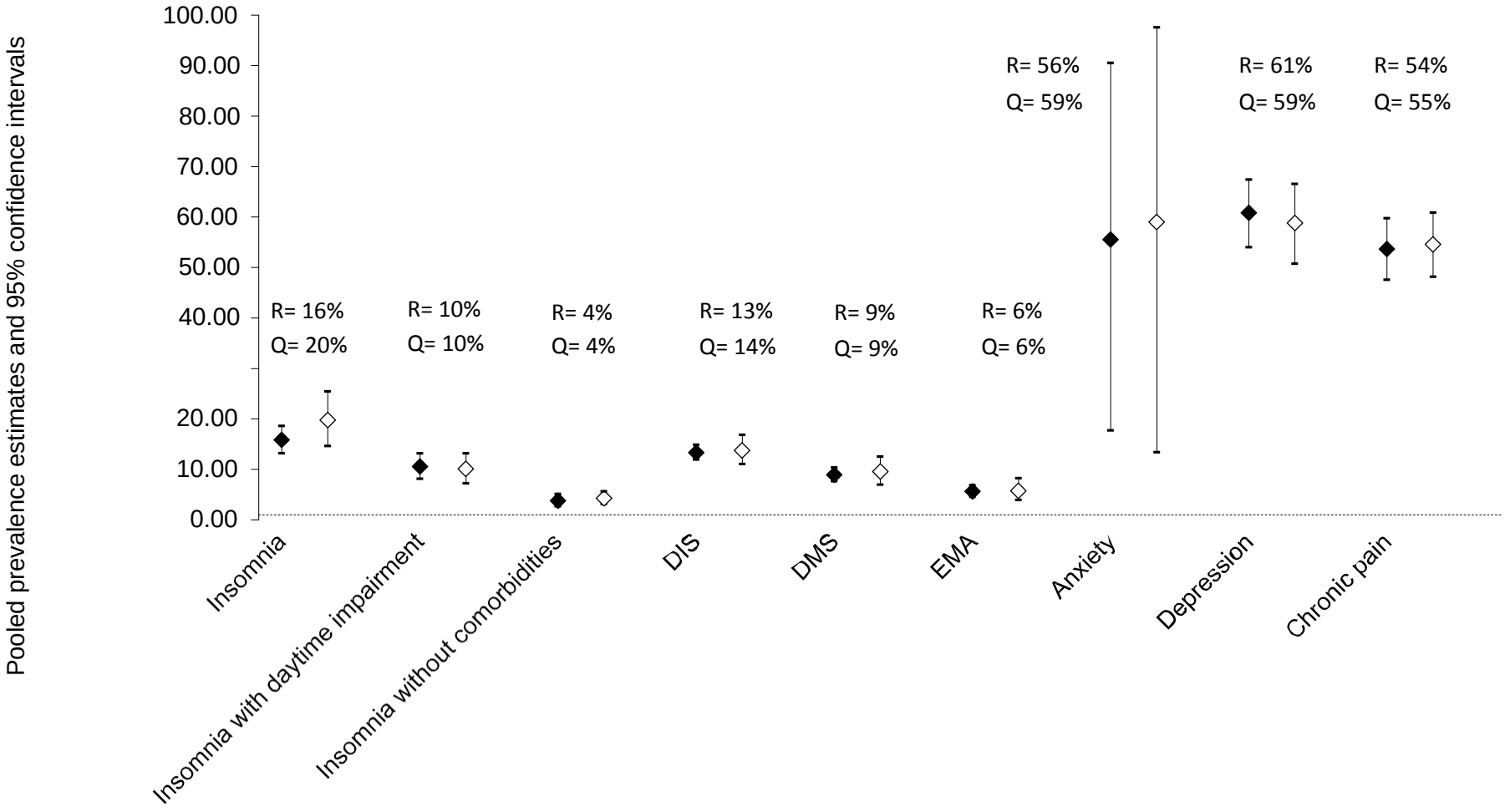

Figure 3.1 - Pooled prevalence estimates and 95% confidence intervals for each definition of insomnia in the non-clinical sample, and for each clinical sample. R= Random-effects; Q= Quality-effects;◆ = Random-effects;◇ = Quality-effects.

3.1.4 Application

This is the first meta-analysis of the prevalence of insomnia in the child and adolescent population, and provides not only a synthesised summary of the prevalence estimates in the empirical literature, but also an overview of the strengths and limitations of existing research. It was observed that the prevalence estimates reported by the articles in the meta-analyses were very discrepant, and it is suggested that this is due to the wide variety of definitions of insomnia, and the methodology used by authors. Future research should seek to define and assess the construct of insomnia carefully, or be clear about the limitations surrounding the generalisability of any findings, particularly with regards to epidemiology or development of intervention. The significant reduction in the prevalence estimates when only including cases of insomnia in the presence of daytime consequences also emphasises the question of which symptoms are clinically relevant for diagnostic purposes, and whether a child experiencing sleep problems in the absence of other symptomatology is any less in need of clinical input.

3.2. Empirical Paper

3.2.1 Background

Autism Spectrum Disorder (ASD) is a pervasive developmental disorder characterised by impairments in social interaction and communication, and restricted or repetitive behaviours or interests (Diagnostic and Statistical Manual: 5th edition; American Psychiatric Association, 2013). Sleep problems have been reported frequently in children with ASD, with consistent evidence of an association between sleep problems and daytime challenging behaviour (Goldman et al., 2011; Malow et al., 2006; Matson, Ancona & Wilkins, 2008; Mazurek & Sohl, 2016;

Schreck, Mulick & Smith, 2004; Tudor, Hoffman & Sweeney (2012). However, the majority of studies employ a cross-sectional design, and compare behaviour between those above and below a sleep disorder cut-off, or explore associations between measures of sleep and behaviour. Thus, the findings could be interpreted as sleep affecting daytime behaviour, as well as daytime behaviour having an impact on sleep. Alternatively, it may be that an underlying factor exists which influences both of these variables. The lack of longitudinal data is a key limitation of existing literature, as the direction of any temporal association between sleep and daytime presentation cannot be identified. Therefore, the aim of this research was to explore the temporal association between sleep and daytime challenging behaviour.

3.2.2 Methodology

Seventeen children with ASD, aged 5-13 years (mean= 10.1, SD= 2.2), and their families completed questionnaire assessments of both sleep problems and challenging behaviour, as well as participating in a seven-day sleep study. During the sleep study, actigraphy data were collected alongside sleep diaries and twice-daily ratings of challenging behaviour and irritability. Actigraphy is a method used to estimate sleep duration and quality, based on the wearer's movement and the presence of white light. The data is then entered into a software package, which uses this information to calculate the times during which the child is asleep, and to generate statistics such as sleep efficiency, total sleep time, and sleep onset latency[1].

[1] Sleep efficiency is an estimate of sleep quality, based on the time a child has spent asleep, in relation to the time they have been in bed, and the period of awakenings during the night. Sleep onset latency refers to the time taken for a child to fall asleep after lights out.

3.2.3 Results

Eighty-seven per-cent of the sample had scores indicative of a sleep problem. The study showed that the children who had the highest scores for sleep problems also had the highest scores on measures of challenging behaviour. An association was observed between sleep problems and daytime behaviours; specifically, the number of awakenings during the night and overall challenging behaviour score.

Regression analyses using the actigraphy data identified that time spent awake after falling asleep the previous night, interacted with parental ratings of irritability to predict severity ratings of daytime challenging behaviour during the subsequent day. None of the sleep variables predicted subsequent daytime irritability, and neither irritability nor challenging behaviour predicted sleep the following night. This suggests that there is a direction of influence between sleep problems, specifically night-time waking, irritability and daytime challenging behaviour.

3.2.4 Application

The findings support the existing literature, which suggests children with sleep problems are more likely to also present with challenging behaviour. However, the present study is the first to provide evidence for a directional association between sleep problems and subsequent daytime behaviour. This finding therefore has implications for the way in which clinicians assess for the contributors to challenging behaviour, and the way in which interventions are developed.

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Volume I Appendices