



Provided by the author(s) and University of Galway in accordance with publisher policies. Please cite the published version when available.

Title	Familial sleep and autism spectrum disorder: a pilot actigraphy study of sleep quality, quality of life and psychological distress.
Author(s)	Leader, Geraldine; Glynn, Carmel; Kirkpatrick, Bernadette; Chen, June L.; O'Súilleabháin, Páraic S.; Mannion, Arlene
Publication Date	2021-05-11
Publication Information	Leader, Geraldine, Glynn, Carmel, Kirkpatrick, Bernadette, Chen, June L., O'Súilleabháin, Páraic S., & Mannion, Arlene. (2021). Familial sleep and autism spectrum disorder: a pilot actigraphy study of sleep quality, quality of life and psychological distress. <i>Irish Journal of Psychological Medicine</i> , doi:10.1017/ipm.2021.24
Publisher	Cambridge University Press
Link to publisher's version	<a href="https://dx.doi.org/10.1017/ipm.2021.24">https://dx.doi.org/10.1017/ipm.2021.24</a>
Item record	<a href="http://hdl.handle.net/10379/16764">http://hdl.handle.net/10379/16764</a>
DOI	<a href="http://dx.doi.org/10.1017/ipm.2021.24">http://dx.doi.org/10.1017/ipm.2021.24</a>

Downloaded 2024-04-27T05:05:42Z

Some rights reserved. For more information, please see the item record link above.



Running head: FAMILIAL SLEEP AND ASD

This is a post-print of an article published in *Irish Journal of Psychological Medicine*.

The final version is available online at:

doi:10.1017/ipm.2021.24

**Familial sleep and autism spectrum disorder: A pilot actigraphy study of sleep quality, quality of life, and psychological distress**

Geraldine Leader<sup>1</sup>

Carmel Glynn<sup>1</sup>

Bernadette Kirkpatrick<sup>2</sup>

June L. Chen<sup>3</sup>

Páraic S. O'Súilleabháin<sup>4,5</sup>

Arlene Mannion<sup>1</sup>

<sup>1</sup>Irish Centre for Autism and Neurodevelopmental Research (ICAN), School of Psychology, National University of Ireland, Galway.

<sup>2</sup>University of Ulster, Coleraine, Northern Ireland.

<sup>3</sup>Department of Special Education, Faculty of Education, East China Normal University, Shanghai, China.

<sup>4</sup>Department of Psychology, University of Limerick, Limerick, Ireland.

<sup>5</sup>Health Research Institute, University of Limerick, Limerick, Ireland.  
Correspondence: Geraldine Leader, Ph.D., Irish Centre for Autism and

Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland. Tel: 00353 91 493434, Fax: 00353 91 521355.

Corresponding Author Email Address: [geraldine.leader@nuigalway.ie](mailto:geraldine.leader@nuigalway.ie)

## Abstract

**Objectives.** Sleep problems are common among children with autism spectrum disorder (ASD) and can have a negative impact on the child's behaviour and daytime functioning. The current pilot study examined objective measurements of child and parent sleep as factors associated with the stress, anxiety, depressive symptoms, social support, and quality of life of parents of children with ASD.

**Methods.** Participants were nine children with ASD and their parents (nine mothers and three fathers). Participants wore an actigraph for 7 consecutive days and nights. Measures of sleep habits and quality was used to measure child and parent sleep. Measures of parenting stress, anxiety, depressive symptoms, quality of life, and social support were collated.

**Results.** Results indicated the emergence of high parental stress, anxiety and depressive symptoms. Significant correlations were observed between parent depressive symptoms, and both subjective sleep quality and child sleep disruptions.

**Conclusions.** The present study found that parental well-being is affected by child sleep problems.

*Keywords:* autism spectrum disorder, sleep problems, actigraphy, parents, quality of life

## FAMILIAL SLEEP AND ASD

### **Familial sleep and autism spectrum disorder: A pilot actigraphy study of sleep quality, quality of life and psychological distress**

#### **Introduction**

Sleep problems are common in children and adolescents with autism spectrum disorder (ASD) (Richdale & Schreck, 2009; Mannion & Leader, 2013; Wiggs & Stores, 2004). Several studies have reported settling problems with some children with ASD taking more than an hour to fall asleep (Angriman et al., 2015; Cortesi et al., 2010; Kotagal, & Broomall, 2012). The reported prevalence of sleep problems in ASD has varied in the research from 33% (Goldman et al., 2011) to 80% (Mannion et al. (2013). Sleep problems have been found to persist over time in children and adolescents with ASD (Mannion & Leader, 2016). Prevalence of sleep problems in children with ASD is much higher than in typically developing children (Souders et al., 2009), with research finding sleep problems were more than twice as common in children with ASD as in the general population (Reynolds et al., 2019).

#### **Impact of sleep problems on the child**

Sleep problems in children with ASD have a negative impact on various other aspects of the child's life. Many children with ASD and other developmental disorders experience other comorbid conditions such as behaviour problems, toileting problems, attention-deficit/hyperactivity disorder (AD/HD), epilepsy, and gastrointestinal symptoms (Devlin et al., 2008; Francis et al., 2017; Leader et al., 2018; Leader & Mannion, 2016a; 2016b; Mannion & Leader, 2014a; 2014b; Newman et al., 2015). Research has begun to investigate the relationships between sleep problems and other comorbid conditions in ASD.

Positive correlations have been found between sleep problems and daytime challenging behaviour (Adams et al., 2014; Goldman et al., 2011; Sikora et al., 2012;

## FAMILIAL SLEEP AND ASD

Tudor et al., 2012). Correlations have also been found between sleep problems and hyperactivity, and compulsive and ritualistic behaviours, compared to children without sleep problems (Goldman et al., 2009). Research has found significant positive associations between sleep problems, anxiety, and challenging behaviour, with medication, anxiety and sleep problems accounting for 42% of the variance in challenging behaviour (Rzepecka et al., 2011). Sleep problems have been shown to be a predictor of anxiety in children and adolescents with ASD (Williams et al., 2015).

Research has found that fewer hours of sleep per night predicted overall autism severity scores and social skills deficits (Adams et al., 2014; Schreck et al., 2004). Stereotypic behaviour was predicted by fewer hours of sleep per night and screaming during the night, and poor sleepers with ASD have a higher percentage of behavioural problems than good sleepers with ASD (Schreck et al., 2004).

### **Impact of sleep problems on parents**

Sleep problems that affect children with ASD also affect parent's sleep quality and quantity. Children waking during the night usually need parental help and/or attention, resulting in sleep loss for one or both parents (McCann et al., 2015; Meltzer & Mindell, 2007). Parent sleep can be affected by other factors, such as stress. Parents of children with developmental disabilities reported poorer sleep quality than parents of typically developing children, and that the strongest predictor of poor sleep quality was parental stress (Gallagher et al., 2010).

Research has found that children's sleep problems have a negative impact on parent anxiety and depressive symptoms levels. Children's sleep behaviours have been shown to place a strain on marital relationships and financial insecurity (Kirkpatrick et al., 2019). Research has shown that maternal sleep quality was a

## FAMILIAL SLEEP AND ASD

significant predictor of maternal mood, stress, and fatigue (Bar et al., 2016; Kouros & El-Sheikh, 2016; Meltzer & Mindell, 2007). Children with ASD who had significant sleep disruptions were significantly more likely to have mothers with more depressive symptoms, fatigue, and daytime sleepiness (Delahaye et al., 2014; Hodge et al., 2013). Mothers of children who had significant sleep disruptions reported greater feelings of parenting stress and care giving overload. Greater sleep and behaviour problems in children had a significant association with disturbed sleep and increased depressive symptoms, anxiety and stress levels in mothers (Chu & Richdale, 2009).

Children's sleep significantly predicted maternal mental health, maternal sleep and maternal stress, and maternal sleep and maternal stress each predicted maternal mental health (Hodge et al., 2013). Children's sleep and maternal sleep were more closely related to maternal stress for mothers of typically developing children, and it was concluded that this may be because stress related to sleep is minor relative to other sources of stress relating to their child's disability (Hodge et al., 2013).

### **Actigraph for monitoring and tracking sleep.**

Strong evidence would support the use of the actigraph to objectively monitor and track sleep (Adkins et al., 2012; Meltzer et al., 2012; Wiggs & Stores, 2004). The actigraph device can be used at home and is considered a reliable and an objective method for measuring sleep-wake patterns, onset of sleep, continuity, and sleep duration (Wiggs & Stores, 2004), making the actigraph suitable for the current study.

### **Current study**

The current pilot study aimed to expand on existing research in critical ways. We primarily sought to extend current literature by using objective sleep measurements through the use of actigraphy. In doing so, we sought to assess sleep in children with ASD and their parents. We also examined subjective measures of sleep,

## FAMILIAL SLEEP AND ASD

including further measures associated with wellbeing, namely parental stress, anxiety, depressive symptoms, and overall quality of life. This pilot study sought to address several limitations in existing research at this critical research intersection, while providing a basis for future research in this area.

### **Method**

#### **Participants**

Families of children with ASD were recruited for the current study if they had a child aged between 3 and 18 years and were recruited through special schools, schools with ASD units, and from parent support groups. Once an interested individual contacted the researchers, and it was confirmed that their child had an independent diagnosis of ASD, and were aged between 3 and 18 years, they were recruited for the study.

The study sample comprised of nine children and adolescents with a diagnosis of ASD in accordance with DSM-IV-TR criteria (American Psychiatric Association, 2000). Diagnoses were provided by a licenced psychologist or paediatrician independent of the study. The participants received their diagnosis as a result of the formal diagnostic protocol which employs multiple diagnostic measures. Caregiver information on professional diagnosis, diagnostic setting/organization and professional(s) who made the diagnosis was obtained.

The final sample consisted of a total of 21 participants (nine families – nine mothers, three fathers and nine children). Selection bias may have occurred. We attempted to address this by advertising that we were looking to recruit families where the child did not have sleep problems also. However, it is possible that only families where the child had sleep problems chose to participate in the study. Some children

## FAMILIAL SLEEP AND ASD

were not able to tolerate the use of actigraphs, which impacted upon the attrition rate.

The descriptive statistics for participants are presented in Table 1.

---Insert Table 1 about here---

### Measures

The questionnaire booklet consisted of a demographic questionnaire, the below questionnaires and sleep diaries for the parent(s) and child. The demographic questionnaire consisted of questions on the child's age, gender, diagnosis, co-morbidity, education, and medication.

**Sleep diary.** Participants completed a sleep diary for 7 days, for themselves and their children. They recorded when the actigraph was removed, and what time it was replaced, the start and end time of any naps taken, time got into bed and time turned out lights, time woke up in the morning, time turned on lights and time got out of bed. Research has suggested that it is useful to use a sleep diary, as it has been shown to be an important adjunct to actigraphy when used to edit data and remove artefacts (Wiggs & Stores, 2004).

**The Pittsburgh Sleep Quality Index (PSQI).** The participants (mothers and fathers) completed the PSQI (Buysse et al., 1989). The 19-item self-report questionnaire assesses sleep quality and sleep disruption. Seven dimensions of sleep: subjective sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbances, use of sleeping medications, and daytime dysfunction were measured. The component scores are summed to give a total sleep quality score, which ranges from 0 to 21. A total score of  $\geq 5$  has been recommended as clinically significant in terms of poor sleep quality. The scale has acceptable internal consistency and good test-retest reliability (Buysse et al., 1989).

## FAMILIAL SLEEP AND ASD

**The Parenting Stress Index-Short Form (PSI/SF-4).** The PSI/SF-4 (Abidin, 2012) is a standardised measure used to evaluate stress in parents that are at risk for parenting problems or child behavioural problems. The questionnaire consists of 36 items pertaining to parental feelings and experiences, comprising three scales: Parental Distress, Parent–Child Dysfunctional Interaction, and Difficult Child Characteristics. Parents rate their agreement for each statement on a 5-point Likert scale, ranging from 1 (strongly agree) to 5 (strongly disagree). The scales have shown high internal consistency and adequate test–retest reliability and have been used widely in studies of parents of children with disabilities (Dardas & Ahmad, 2014).

**Multidimensional Scale of Perceived Social Support (MSPSS).** The MSPSS (Zimet et al., 1988) is a 12-item self-report measure that evaluates a person’s perceived social support, from three subscales – family, friends and significant other. It was utilised as an additional variable to analyse parent stress. The MSPSS has demonstrated good internal and test-retest reliability. With high levels of social support associated with low levels of depressive symptoms and anxiety (Canty-Mitchell & Zimet, 2000; Zimet et al., 1988).

**The Hospital Anxiety and Depression Scale (HADS).** The HADS (Zigmond & Snaith, 1983) was used to assess parent levels of anxiety and depressive symptoms. The scale consists of 14 items, seven assessing anxiety and seven assessing depressive symptoms. It yields a total depressive symptoms score and a total anxiety score. A score of 0-7 is classified as normal for each score, 8-10 is classified as borderline abnormal (borderline case) and 11-21 is classified as abnormal. It has demonstrated good internal validity and concurrent validity, and has been found to perform well in assessing the symptom severity and cases of anxiety

## FAMILIAL SLEEP AND ASD

disorders and depressive symptoms in somatic, psychiatric and primary care patients and in the general population (Bjellend et al., 2002).

**WHOQOL-BREF (World Health Organization's Quality of Life Questionnaire-BREF).** Parent's quality of life was assessed using the WHOQOL-BREF (World Health Organization; The WHOQOL Group; 1998), an abbreviated version of the WHOQOL-100. The questionnaire consists of several demographic questions at the beginning, followed by 26 items which are rated on a 5-point scale. These items assess four domains: physical health, psychological, social relationships and environment. The mean score of items within each domain is used to calculate the domain score. The scale has been shown to have good reliability and validity in adult populations (Krägeloh et al., 2013).

**Actigraphy.** All participants (mothers, fathers and children) were asked to wear an actigraph (Actiwatch 2, Philips Respironics, Norfolk, UK) for 7 consecutive days and nights, worn on the non-dominant wrist. The actigraphs were configured by using a 1-minute epoch, with medium threshold, as this is the most widely used epoch length in sleep research (Meltzer et al., 2012). Parents were instructed to press the event marker button on the side of the actigraph to mark sleep onset and sleep offset on their own actigraphs, and to do so on their child's actigraph also. Objective sleep quantity was measured by total sleep time (TST, total time within rest intervals scored as sleep). Additional data gathered and their definitions are as follows. (1) bed time (time when the subject went to bed with the intent to sleep). (2) get up time (time when the subject rose from bed for the final time). (3) time in bed (period of time between bed time and get up time). (4) onset latency (period of time between bed time and sleep start time). (5) sleep efficiency (percentage of time in bed actually spent sleeping). (6) Wake After Sleep Onset (WASO; the number of waking minutes

## FAMILIAL SLEEP AND ASD

between sleep start and sleep end times) and (7) #Awak (the number of continuous blocks of wake). A validated Philips Actiware software (Version 6, Philips Respironics) algorithm was used to estimate sleep parameters, based on thresholds for wake and sleep (Adkins et al., 2012). Actigraphy has been widely used in recent years in sleep studies with both children and adults, and has demonstrated good reliability and validity (Sadeh, 2011).

### **Procedure**

A home visit was arranged at a time convenient for the family. Data collection took place during the summer holidays. Informed consent was obtained from the parents, and verbal assent from the children. Parents were then given the actigraphs and shown how to use them and given instructions on proper care of them. They were also instructed on the use of the daily sleep diary. They were also given the questionnaire booklet, containing the questionnaires listed above. After 7 days, a second home visit was made to collect the actigraphs, sleep diaries and questionnaires.

### **Results**

#### **Parenting Stress Index-SF (PSI-SF)**

The majority of parents scored highly on the PSI-SF, indicating high parenting related stress. The means, standard deviations, range, minimum and maximum scores are presented in Table 2 below.

---Insert Table about 2 here---

#### **Multidimensional Scale of Perceived Social Support (MSPSS)**

## FAMILIAL SLEEP AND ASD

The majority of participants scored highly on the MSPSS. The means, standard deviations, range, minimum and maximum scores are presented in Table 2 above.

### **Hospital Anxiety and Depression Scale (HADS)**

The majority of participants scored highly on both the anxiety and depressive symptoms subscales of the HADS. The means, standard deviations, range, minimum and maximum scores are presented in Table 2 above.

### **WHOQOL-BREF**

The raw scores calculated for each of the four domains were transformed into a 0-100 scale score. Lower scores indicate lower quality of life. The majority of participants had lower than average quality of life scores. Hawthorne et al. (2006) investigated population normative values for the WHOQOL-BREF. The normative values they found were 73.5 for Domain 1, 70.6 for Domain 2, 71.5 for Domain 3 and 75.1 for Domain 4, with an average for the four subscales of 72.66. The means, standard deviations, range, minimum and maximum scores are presented in Table 2 above.

### **Subjective Sleep Quality**

The mean PSQI total score was higher for mothers ( $M= 8.56$ ,  $SD= 3.36$ ) than for fathers ( $M= 7.00$ ,  $SD= 4.00$ ), with a high mean total score for parents ( $M= 8.17$ ,  $SD= 3.41$ ). Meltzer (2008) stated that a total score of  $\geq 5$  on the PSQI is recommended as clinically significant in terms of poor sleep quality. Two parents (17%) had a total score of  $< 5$  on the PSQI, while 10 parents (83%) had a total score of  $\geq 5$ , indicating that the majority of parents had self-reported sleep problems. The means, standard deviations and range of scores are presented in Table 3 below.

---Insert Table 3 here---

## FAMILIAL SLEEP AND ASD

### **Actigraphy**

A week (7 days and nights) of actigraph measurement produced an average for each of the sleep variables for both parents and children. The means and standard deviations for the range of sleep variables for parents are presented in Table 4. Souders et al. (2009) stated that the Children's Hospital of Philadelphia's (CHOP) Sleep Centre recommends a sleep efficiency of  $<85\%$  and a sleep onset latency of  $>30$  minutes as a cut-off for defining poor sleep quality. The majority of participants did not have poor sleep quality as indicated by poor sleep efficiency, with three participants (25%) having a sleep efficiency of  $<85\%$ , while nine participants (75%) had a sleep efficiency of  $\geq 85\%$ . Total sleep time was less than recommended, with nine participants (75%) having  $\leq 7$  hours of sleep, with three participants (25%) having  $>7$  hours.

---Insert Table 4 about here---

The means and standard deviations for the range of sleep variables for children were included in Table 5. Eight children (89%) had a total sleep time of  $\leq 8$  hours, with one child (11%) getting  $>8$  hours sleep. Four children (44%) had  $\leq 7$  hours of sleep, with five children having  $>7$  hours of sleep per night, indicating less than recommended sleep time. Five children (56%) had  $<85\%$  sleep efficiency, while four children (44%) had  $>85\%$  sleep efficiency. The mean sleep efficiency of 83% is lower than the recommended cut-off of 85% indicating poor sleep quality.

---Insert Table 5 about here---

### **Correlation Analyses**

**Subjective and objective sleep measures.** Pearson's correlations were used to test for associations between the subjective and objective sleep variables. No

## FAMILIAL SLEEP AND ASD

significant relationships were found between the subjective sleep measures (PSQI) and any of the objective (actigraph) sleep variables.

**Sleep issues and stress.** Pearson's correlations were conducted to determine if there was a relationship between self-reported sleep quality (PSQI total score) and parent stress (PSI total stress), and between actigraph variables and parent stress. There were no significant correlations between parent stress and subjective sleep quality, or between parent stress and any of the child actigraph variables. There was only one significant correlation for the parent actigraph variables, a positive association between PSI total stress and sleep efficiency (as measured by actigraph), ( $r(12) = .61, p = .04$ ), with a large effect size.

**Sleep issues and parent depressive symptoms.** Pearson's correlations were conducted to determine if there was a relationship between self-reported sleep quality and parent depressive symptoms, and between actigraph variables and parent depressive symptoms. A large positive correlation was found between the global PSQI score and HADS depressive symptoms score, ( $r(12) = .70, p < .05$ ). A significant negative correlation was found between HADS depressive symptoms score and parent sleep latency, ( $r(12) = -.70, p = .01$ ), with a large effect size. Of the child actigraph variables, one significant correlation was found, between child wake after sleep onset and HADS depressive symptoms score, ( $r(12) = .63, p = .03$ ), with a large effect size.

**Sleep issues and parent anxiety.** Pearson's correlations were conducted to determine if there was a relationship between self-reported sleep quality and parent anxiety, and between actigraph variables and parent anxiety, with no significant correlations found.

## FAMILIAL SLEEP AND ASD

**Sleep issues and quality of life.** Pearson's correlations were conducted to determine if there was a relationship between self-reported sleep quality and parent quality of life, and between actigraph variables and parent quality of life. One significant positive correlation was found between average WHOQOL-BREF scores and sleep efficiency (actigraphy), ( $r(12) = .60, p = .04$ , with a large effect size).

**Parent sleep variables and child sleep variables.** Pearson's correlations were conducted to determine if there was a relationship between parent actigraph variables and child actigraph variables, with no significant correlations found.

### Discussion

The current pilot study was the first study to use a multi-method approach to examine both child and parent sleep, and their relationship with parent stress, anxiety, depressive symptoms, and quality of life. The current study diverges from previous findings that report child sleep disruption affects parent sleep, in that within the current sample child shorter total sleep time, lower sleep efficiency and waking after sleep onset did not impact on parent sleep. One possible reason is that the child may not have sought parental attention, which is a factor which may warrant further investigation.

A significant association was found between parent depressive symptoms and self-reported subjective sleep quality, consistent with previous research (Chu & Richdale, 2009; Hall et al., 2017; Meltzer, 2011). A significant positive association was found between parent depressive symptoms and child wake after sleep onset, i.e. that higher parent depressive symptoms was associated with child sleep disruptions during the night. In addition to depressive symptoms, research has indicated that sleep disturbance and sleep disorders could be linked to alcohol use and abuse in alcoholics and non-alcoholics (Roehrs et al., 2001). Caffeine consumption has also

## FAMILIAL SLEEP AND ASD

been reported as having a detrimental impact on sleep patterns especially in adolescents (Snel et al., 2011). Evidence also suggests that nicotine influences sleep and mood and that smoking increased sleep related respiratory disorders which negatively impact on sleep quality and increased day time tiredness (Jaehne et al., 2009). It is possible that parent self-reported reduced quantity and good quality of sleep could have been linked to alcohol use or abuse, caffeine and or nicotine consumption. Although beyond the scope of the current study, the measurement of the aforementioned sleep problem contributors would be considered beneficial to future research.

A positive correlation was found between quality of life and parent sleep efficiency, i.e. lower quality of life scores was associated with lower sleep efficiency. This may be because individuals with a lower quality of life have many worries, e.g. money, work, children, lack of social support, etc., which impact on their sleep quality. Although lower quality of life may not have an impact physically, it has an impact on psychological well-being, therefore contributing to the poorer sleep quality.

A significant negative association was found between parent depressive symptoms and parent sleep onset latency, indicating that higher parent depressive symptoms was associated with shorter sleep onset latency. No significant correlations were found between the subjective sleep measures (PSQI) and the objective sleep measures (actigraphy). This may suggest that subjective measures of sleep may be unreliable compared to objective measures such as actigraphy, and that self-report measures may over estimate or exaggerate sleep problems. It may also suggest that parents feel frustrated or burnt out. The subjective and objective measures of sleep did not match well, which indicates that subjective measures of sleep should be

## FAMILIAL SLEEP AND ASD

improved in the future. The concordance between subjective sleep measures and actigraphy should be addressed in future research.

The main findings of the study indicate that parental depressive symptoms may be linked to self-reported sleep quality, and child sleep disruptions. Parental self-reported depressive symptoms were higher than average normative values (Hinz & Brähler, 2011), as was self-reported anxiety. No associations were found between parent anxiety and parent or child sleep, which may suggest that parental anxiety is linked to other child issues, e.g. child behaviour problems, symptom severity, etc. Certain sleep issues were apparent in the children in this study, including total sleep times that are less than recommended for children, with 89% of the children in the study getting less than 8 hours of sleep on average per night, and 44% getting less than 7 hours per night. Sleep recommendations indicate that children aged between 7 and 12 years of age should get 10 to 11 hours of sleep per night, and that children aged 12-18 years should get 8 to 9 hours of sleep (National Sleep Foundation, 2015). The average sleep efficiency was lower than the recommended cut-off of 85% (Souders et al., 2009), indicating less than recommended sleep quality. Wake after sleep onset was also an issue for children.

The sleep patterns of parents in this study did not appear abnormal. Bed-times, get up times and total sleep times were wide ranging. Sleep onset latency was short, with an average latency of 9 minutes, and average sleep efficiency was 85%, indicating that the sleep quality was quite good. The high PSQI scores did not appear to correlate with the actigraph variables, apart from less than recommended total sleep time, with parents getting an average of 6 hours 35 minutes of sleep per night (7-8 hours is recommended for adults), and wake after sleep onset was quite high, with

## FAMILIAL SLEEP AND ASD

parents spending an average of 44 minutes awake each night, with 92% of parents awake for longer than 30 minutes each night.

Results were found that also contradict previous research on parent stress and sleep, with a positive correlation found between parent stress and parent sleep efficiency, indicating that higher parent stress was linked to greater parent sleep efficiency. The results indicated that parents with high stress and/or depressive symptoms levels may be more tired, and sleep better as a result. This result is in contrast to previous results (Gallagher et al., 2010), which found that parental stress was associated with poorer sleep quality. However, this result was based on subjective sleep measures, which may not be as reliable as actigraphy. Similar results have been found as the current study, where child and parent sleep were more closely linked to parent stress for parents of typically developing children than for parents of children with ASD (Hodge et al., 2013). This may be because stress related to sleep is minor relative to other sources of stress relating to their child's disability.

Parent stress, anxiety and depressive symptoms were all higher than average, with quality of life lower than average. The mean PSI stress score was in the 86<sup>th</sup> percentile, indicating very high stress levels for parents. As no associations were found between parent stress and parent or child sleep problems, it would appear that parent stress may be linked to other factors not examined in the current study, for example, child behaviour or child functioning. Furthermore, 89% of mothers and 67% of fathers scored above average for both self-reported anxiety and depressive symptoms. In support of this, previous research has found that 83% of parents reported lower than average quality of life (Hawthorne et al., 2006), which again was not found to be linked to parent or child sleep problems.

## FAMILIAL SLEEP AND ASD

The current study had several methodological strengths, including objective measurement (actigraphy) of both parent and child sleep, which has been used in very few studies. The questionnaires used in the study have been validated and shown to be reliable. The study examined several relationships between parent sleep and parent issues, and child sleep and parent issues.

There were also several limitations. Firstly, this was a very small study, where the small sample size limited the statistical analyses that could be conducted on the data, as well as the generality and validity of the study. The failure to find relationships could be due to several uncontrolled factors, or to the small sample size. Non-tolerance of the actigraphs was a major issue in this study, with several participants having to pull out due to being unable to tolerate the actigraphs. Tactile sensitivity is a major issue for many children with ASD; therefore, asking a child to wear a watch-like device and can be difficult for many children. Data collection took place during the summer holidays, which may have affected the availability of families to participate in the research. The absence of a control group is also a limitation of the present study. A control group would have allowed comparisons to be drawn between the groups for each of the variables examined. A final limitation was the lack of fathers who participated in the study, with only three out of the nine families having both mother and father. This made it difficult to compare maternal data with paternal data due to the limited numbers.

The present study raises important points for future research. No correlations were found between subjective and objective measures of sleep, highlighting the lack of reliability of subjective sleep measures, which can over exaggerate sleep problems. There was a lack of relationships found between parent stress, anxiety, depressive symptoms and quality of life and child and parent sleep in the current study; therefore,

## FAMILIAL SLEEP AND ASD

the factors that affect these variables should be examined in future research.

Actigraphy is an emerging and more reliable method of sleep measurement. A multi-method approach to sleep measurement, which allows an examination of different aspects of sleep, should be used in all future sleep research. A replication of this study with a larger sample size and a typically developing control group should be conducted in the future.

## FAMILIAL SLEEP AND ASD

**Conflicts of interest:** None.

### **Ethical Standards**

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committee on human experimentation with the Helsinki Declaration of 1975, as revised in 2008. Informed consent was obtained from all individual participants included in the study. The study protocol was approved by the ethics committee of the participating institution.

### **Financial Support**

This research received no specific grant from any funding agency, commercial or not-for-profit sectors.

### References

- Abidin, R.R.** (2012). *Parenting Stress Index* (4th ed). Odessa, FL: Psychological Assessment Resource.
- Adams, H. L., Matson, J. L., Cervantes, P. E., & Goldin, R. L.** (2014). The relationship between autism symptom severity and sleep problems: Should bidirectionality be considered? *Research in Autism Spectrum Disorders, 8*, 193-199.
- Adkins, K. W., Molloy, C., Weiss, S. K., Reynolds, A., Goldman, S. E., Burnette, C., ... & Malow, B. A.** (2012). Effects of a standardized pamphlet on insomnia in children with autism spectrum disorders. *Pediatrics, 130*, S139-S144.
- American Psychiatric Association** (2000). *Diagnostic and statistical manual of mental disorders (4th ed.) text revision (DSM-IV-TR)*. American Psychiatric Association, Arlington, VA.
- Angriman, M., Caravale, B., Novelli, L., Ferri, R., & Bruni, O.** (2015). Sleep in children with neurodevelopmental disabilities. *Neuropediatrics, 46*, 199-210.
- Bar, M., Efron, M., Gothelf, D., & Kushnir, J.** (2016). The link between parent and child sleep disturbances in children with attention deficit/hyperactivity disorder. *Sleep medicine, 21*, 160-164.
- Bjellend, I., Dahl, A.A., Haug, T.T., & Neckelmann, D.** (2002). The validity of the hospital anxiety and depression scale: an updated literature review. *Journal of Psychosomatic Research, 52*, 69-77.
- Buyse, D.J., Reynolds, C.F., Monk, T.H., Berman, S.R., & Kupfer, D.J.** (1989).

## FAMILIAL SLEEP AND ASD

- The Pittsburgh sleep quality index: a new instrument for psychiatric practice and research. *Psychiatry Research*, **28**, 198-213.
- Canty-Mitchell, J. & Zimet, G.D.** (2000). Psychometric properties of the Multidimensional Scale of Perceived Social Support in urban adolescents. *American Journal of Community Psychology*, **28**, 391-400.
- Chu, J., & Richdale, A.L.** (2009). Sleep quality and psychological wellbeing in mothers of children with developmental disabilities. *Research in Developmental Disabilities*, **30**, 1512-1522.
- Cortesi, F., Giannotti, F., Ivanenko, A., & Johnson, K.** (2010). Sleep in children with autistic spectrum disorder. *Sleep Medicine*, **11**, 659-664.
- Dardas, L.A., & Ahmad, M.M.** (2014). Psychometric properties of the Parenting Stress Index with parents of children with autistic disorder. *Journal of Intellectual Disability Research*, **58**, 560-571.
- Delahaye, J., Kovacs, E., Sikora, D., Hall, T. A., Orlich, F., Clemons, T. E., & Kuhlthau, K.** (2014). The relationship between Health-Related Quality of Life and sleep problems in children with Autism Spectrum Disorders. *Research in Autism Spectrum Disorders*, **8**, 292–303.
- Devlin, S., Healy, O., Leader, G., & Reed, P.** (2008). The analysis and treatment of problem behavior evoked by auditory stimulation. *Research in Autism Spectrum Disorders*, **2**, 671-680.
- Francis, K., Mannion, A., & Leader, G.** (2017). The assessment and treatment of toileting difficulties in individuals with autism spectrum disorder and other developmental disabilities. *Review Journal of Autism and Developmental Disorders*, **4**, 190-204.
- Gallagher, S., Phillips, A.C., & Carroll, D.** (2010). Parental stress is associated with

## FAMILIAL SLEEP AND ASD

poor sleep quality in parents caring for children with developmental disabilities. *Journal of Pediatric Psychology*, **35**, 728-737.

**Goldman, S.E., McGrew, S., Johnson, K.P., Richdale, A.L., Clemons, T., & Malow, B.A.** (2011). Sleep is associated with problem behaviours in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders*, **5**, 1223-1229.

**Goldman, S.E., Surdyka, K., Cuevas, R., Adkins, K., Wang, L., & Malow, B.A.** (2009). Defining the sleep phenotype in children with autism. *Developmental Neuropsychology*, **34**, 560-573.

**Hall, W. A., Moynihan, M., Bhagat, R., & Wooldridge, J.** (2017).

Relationships between parental sleep quality, fatigue, cognitions about infant sleep, and parental depression pre and post-intervention for infant behavioral sleep problems. *BMC Pregnancy and Childbirth*, **17**, 104.

**Hawthorne, G., Herrman, H., & Murphy, B.** (2006). Interpreting the WHOQOL BREF: preliminary population norms and effect sizes. *Social Indicators Research*, **77**, 37-59.

**Hinz, A., & Brähler, E.** (2011). Normative values for the Hospital Anxiety and Depression Scale (HADS) in the general German population. *Journal of Psychosomatic Research*, **71**, 74-78.

**Hodge, D., Hoffman, C.D., Sweeney, D.P., & Riggs, M.L.** (2013). Relationship between children's sleep and mental health in mothers of children with and without autism. *Journal of Autism and Developmental Disorders*, **43**, 956-963.

**Jaehne, A., Loessl, B., Bárkai, Z., Riemann, D., & Hornyak, M.** (2009). Effects of nicotine on sleep during consumption, withdrawal and replacement therapy. *Sleep medicine reviews*, **13**, 363-377.

## FAMILIAL SLEEP AND ASD

- Kelly, M. P., Leader, G., & Reed, P.** (2015). Stimulus over-selectivity and extinction-induced recovery of performance as a product of intellectual impairment and autism severity. *Journal of Autism and Developmental Disorders*, **45**, 3098-3106.
- Kirkpatrick, B., Gilroy, S.P., & Leader, G.** (2019). Qualitative study on parents' perspectives of the familial impact of living with a child with autism spectrum disorder who experiences insomnia. *Sleep Medicine*, **62**, 59-68.
- Kouros, C. D., & El-Sheikh, M.** (2017). Within-Family Relations in Objective Sleep Duration, Quality, and Schedule. *Child Development*, **88**, 1983-2000.
- Kotagal, S., & Broomall, E.** (2012). Sleep in children with autism spectrum disorder. *Pediatric neurology*, **47**, 242-251.
- Krägeloh, C. U., Kersten, P., Billington, D. R., Hsu, P. H. C., Shepherd, D., Landon, J., & Feng, X. J.** (2013). Validation of the WHOQOL-BREF quality of life questionnaire for general use in New Zealand: Confirmatory factor analysis and Rasch analysis. *Quality of Life Research*, **22**, 1451-1457.
- Leader, G., Francis, K., Mannion, A., & Chen, J.** (2018). Toileting problems in children and adolescents with parent-reported diagnoses of autism spectrum disorder. *Journal of Developmental and Physical Disabilities*, **30**, 307-327.
- Leader, G., & Mannion, A.** (2016a). Challenging behaviors. In J.L. Matson (ed.), *Handbook of assessment and diagnosis of autism spectrum disorder* (pp. 209-232). Cham: Springer.
- Leader, G., & Mannion, A.** (2016b). Gastrointestinal Disorders. In J.L. Matson (Ed.), *Comorbid Conditions Among Children with Autism Spectrum Disorders* (pp.257-281). Cham: Springer.

## FAMILIAL SLEEP AND ASD

- Mannion, A., & Leader, G.** (2013). An analysis of the predictors of comorbid psychopathology, gastrointestinal symptoms and epilepsy in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, **7**, 1663-1671.
- Mannion, A., & Leader, G.** (2016). An investigation of comorbid psychological disorders, sleep problems, gastrointestinal symptoms and epilepsy in children and adolescents with autism spectrum disorder: A two year follow-up. *Research in Autism Spectrum Disorders*, **22**, 20-33.
- Mannion, A., Leader, G., & Healy, O.** (2013). An investigation of comorbid psychological disorders, sleep problems, gastrointestinal symptoms and epilepsy in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, **7**, 35-42.
- Mannion, A., & Leader, G.** (2014a). Attention-Deficit/Hyperactivity Disorder in Autism Spectrum Disorder. *Research in Autism Spectrum Disorders*, **8**, 432-439.
- Mannion, A., & Leader, G.** (2014b). Epilepsy in Autism Spectrum Disorder. *Research in Autism Spectrum Disorders*, **8**, 354-361.
- Meltzer, L.J.** (2008). Brief report: sleep in parents of children with autism spectrum disorders. *Journal of Pediatric Psychology*, **33**, 380-386.
- Meltzer, L.J.** (2011). Factors associated with depressive symptoms in parents of children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, **5**, 361-367.
- Meltzer, L.J., & Mindell, J.A.** (2007). Relationship between child sleep disturbances and maternal sleep, mood, and parenting stress: a pilot study. *Journal of Family Psychology*, **21**, 67-73.

## FAMILIAL SLEEP AND ASD

**Meltzer, L.J., Montgomery-Downs, H.E., Insana, S.P., & Walsh, C.M.** (2012).

Use of actigraphy for assessment in pediatric sleep research.

*Sleep Medicine Reviews*, **16**, 463-475.

**McCann, D., Bull, R., & Winzenberg, T.** (2015). Sleep deprivation in parents

caring for children with complex needs at home: a mixed methods systematic review. *Journal of family nursing*, **21**, 86-118.

**National Sleep Foundation** (2015). *How Much Sleep Do We Really Need?* Retrieved

from: <https://sleepfoundation.org/media-center/press-release/national-sleep-foundation-recommends-new-sleep-times>

**Newman, I., Leader, G., Chen, J. L., & Mannion, A.** (2015). An analysis of

challenging behavior, comorbid psychopathology, and Attention-Deficit/Hyperactivity Disorder in Fragile X Syndrome. *Research in developmental disabilities*, **38**, 7-17.

**Roehrs, T., & Roth, T.** (2001). Sleep, sleepiness, sleep disorders and alcohol use and abuse. *Sleep medicine reviews*, **5**, 287-297.

**Reynolds, A. M., Soke, G. N., Sabourin, K. R., Hepburn, S., Katz, T., Wiggins,**

**L. D., ... & Levy, S. E.** (2019). Sleep problems in 2-to 5-year-olds with autism spectrum disorder and other developmental delays. *Pediatrics*, **143**, e20180492.

**Richdale, A. L., & Schreck, K. A.** (2009). Sleep problems in autism spectrum

disorders: prevalence, nature, & possible biopsychosocial aetiologies. *Sleep medicine reviews*, **13**, 403-411.

**Rzepecka, H., McKenzie, K., McClure, I., & Murphy, S.** (2011). Sleep, anxiety

and challenging behaviour in children with intellectual disability and/or autism spectrum disorder. *Research in Developmental Disabilities*, **32**, 2758-2766.

## FAMILIAL SLEEP AND ASD

- Sadeh, A.** (2011). The role and validity of actigraphy in sleep medicine: An update. *Sleep Medicine Reviews*, **15**, 259-267.
- Schreck, K.A., Mulick, J.A., & Smith, A.F.** (2004). Sleep problems as possible predictors of intensified symptoms of autism. *Research in Developmental Disabilities*, **25**, 57-66.
- Sikora, D.M., Johnson, K., Clemons, T., & Katz, T.** (2012). The relationship between sleep problems and daytime behavior in children of different ages with autism spectrum disorders. *Pediatrics*, **130**, 83-90.
- Snel, J., & Lorist, M. M.** (2011). Effects of caffeine on sleep and cognition. *Progress in brain research*, **130**, 395-401.
- Souders, M.C., Mason, T.B., Valladares, O., Bucan, M., Levy, S.E., Mandell, D.S.** (2009). Sleep behaviours and sleep quality in children with autism spectrum disorders. *Sleep*, **32**, 1566-1578
- Tudor, M.E., Hoffman, C.D., & Sweeney, D.P.** (2012). Children with autism: sleep problems and symptom severity. *Focus on Autism and Other Developmental Disabilities*, **27**, 254-262.
- The WHOQOL Group.** Development of the World Health Organization WHOQOL-BREF quality of life assessment. *Psychological Medicine*, **28**, 551-558
- Wiggs, L., & Stores, G.** (2004). Sleep patterns and sleep disorders in children with autistic spectrum disorders: insights using parent report and actigraphy. *Developmental Medicine & Child Neurology*, **46**, 372-380.
- Williams, S., Leader, G., Mannion, A., & Chen, J.** (2015). An investigation of anxiety in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, **10**, 30-40.
- Zigmond, A.S., & Snaith R.P.** (1983). The Hospital Anxiety and Depression Scale.

## FAMILIAL SLEEP AND ASD

*Acta Psychiatr Scand*, **67**, 361–370

**Zimet, G.D., Dahlem, N.W., Zimet, S.G. & Farley, G.K.** (1988). The Multidimensional Scale of Perceived Social Support. *Journal of Personality Assessment*, **52**, 30-41.

## FAMILIAL SLEEP AND ASD

Table 1

*Descriptive statistics of participants*

	<i>M (SD)</i>	<i>n (%)</i>		<i>M (SD)</i>	<i>n (%)</i>
<u><i>Parent</i></u>			<u><i>Child</i></u>		
Age (years)	39.75 (6.57)		Age (years)	9.75	
Age range	31 – 53		Age range	(2.53)	
	years			7-14	
<u><i>Gender</i></u>			<u><i>Gender</i></u>		
Male		3(25%)	Male	years	6(67%)
Female		9(75%)	Female		3(33%)
<u><i>Educational level</i></u>			<u><i>Diagnosis</i></u>		
Primary school		0	Autism		8 (89%)
Secondary school		4(33%)			
Tertiary education		8(67%)	Asperger's Syndrome		1(11%)
<u><i>Marital status</i></u>			<u><i>Type of Education</i></u>		
Single		1(8%)	Applied Behavior Analysis		1(11%)
Married		8(67%)			8(89%)
Divorce		2(17%)	Eclectic		
			Intellectual Disability (ID)		5 (56%)
			Mild ID		4 (44%)
			Moderate ID		1 (11%)
			Epilepsy		1 (11%)
			Taking melatonin		1 (11%)
			ADHD diagnosis		0
			Anxiety diagnosis		0

Note. *SD* = standard deviations appear in the parentheses beside the *M*=mean score.

Table 2

*Descriptive statistics for WHOQOL-BREF, HADS, PSI-SF and MSPSS questionnaires*

## FAMILIAL SLEEP AND ASD

Variable	<i>M (SD)</i>	Range	Minimum	Maximum
<b><u>World Health Organisation's</u></b>				
<b><u>Quality of Life Questionnaire-</u></b>				
<b><u>BREF (WHOQOL-BREF)</u></b>				
WHOQOL-BREF Domain 1 (Physical health)	64.25(17.70)	56	38	94
WHOQOL-BREF Domain 2 (Psychological)	52.00 (18.32)	56	25	81
WHOQOL-BREF Domain 3 (Social relationships)	66.67 (18.84)	69	31	100
WHOQOL-BREF Domain 4 (Environment)	56.33 (15.51)	50	38	88
WHOQOL-BREF Average score	59.81 (13.72)	42.25	40.75	83
<b><u>Hospital Anxiety and Depression</u></b>				
<b><u>Scale</u></b>				
HADS Depression Score	7.83 (3.35)	12	2	14
HADS Anxiety Score	9.92 (3.63)	11	4	15
<b><u>Parenting Stress Index</u></b>				
PSI-SF Parental Distress (PD) subscale	37.25 (8.98)	33	21	54
PSI-SF Parent-Child Dysfunctional Interaction (P-CDI) subscale	31.75 (7.36)	21	22	43

## FAMILIAL SLEEP AND ASD

---

PSI-SF Difficult Child (DC)	41.67 (8.26)	26	28	54
subscale				
PSI-SF Total Stress score	110.67	66	79	145
	(20.91)			
<b><u>Multidimensional Scale of</u></b>				
<b><u>Perceived Social Support</u></b>				
MSPSS Total score	57.20	46	32	78
	(12.78)			

---

## FAMILIAL SLEEP AND ASD

Table 3

*Means, standard deviations and range for subjective sleep variables for parent*

Variable	Mothers ( <i>n</i> =9)	Fathers ( <i>n</i> =3)	Parents total ( <i>N</i> =12)	
	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )	Range
PSQI sleep quality	1.56 (1.01)	1 (1.00)	1.42 (1.00)	
PSQI sleep onset latency	1.33 (1.00)	1.33 (1.16)	1.33 (.99)	
PSQI sleep duration	1.33 (.58)	1.33 (1.00)	1.33 (.89)	
PSQI sleep efficiency	1.00 (1.12)	.67 (.58)	.92 (1.00)	
PSQI sleep disturbances	1.56 (.53)	1.67 (.58)	1.58 (.52)	
PSQI sleep medications	.22 (.67)	.00 (.00)	.17 (.58)	
PSQI daytime functioning	1.56 (.73)	1.00 (1.00)	1.42 (.79)	
PSQI total score	8.56 (3.36)	7.00 (4.00)	8.17 (3.41)	3-14

Note. *SD* = standard deviations appear in the parentheses beside the *M*=mean score.  
 PSQI = Pittsburgh Sleep Quality Index.

## FAMILIAL SLEEP AND ASD

Table 4

*Means, standard deviations and range for objective sleep variables for parents.*

Objective sleep variables (actigraphy)	Mothers ( <i>n</i> =9)	Fathers ( <i>n</i> =3)	Parents total ( <i>n</i> =12)
	<u>Mean (SD)</u>	<u>Mean (SD)</u>	<u>Mean (SD), range</u>
Bedtime	00:55 am (0:46)	01:40 am (2:50)	1:06 am (1:25), range = 23:52 – 4:06)
Get up time	8:48 am (1:34)	8:50 am (2:08)	8:49 am (1:37), range = 6:26 – 11.25)
Time in bed (hours)	7 hrs 53 mins (0:58)	7 hrs 10 mins (0:51)	7 hrs 42 mins (0:57), range = (6 hrs 17 mins – 9 hrs 23 mins)
Total sleep time (hours)	6 hrs 52 mins (0:59)	5 hrs 45 mins (0:11)	6 hrs 35 mins (1:05), range = 4 hrs 55 mins – 8 hrs 22 mins
Onset latency (minutes)	8 mins (0:07)	11 mins (0:11)	9 mins (0:07), range = 1 min – 24 mins
Sleep efficiency (percent)	87% (3.37)	80% (7.16)	85% (5.20), range = 73.28 – 91.14
Wake after sleep onset (minutes)	42 mins (0:13)	50 mins (0:09)	44 mins (0:12), range = 23 mins – 1 hr 1 min
No. of wake intervals	22.44 (5.24)	26.86 (3.95)	23.55 (5.17), range = 15.86 – 31.86

## FAMILIAL SLEEP AND ASD

Table 5

*Means, standard deviations and range for objective sleep variables for children and adolescents with ASD.*

Variable	Mean (standard deviation), range
Bedtime	00:00 am (1:36), 22:07 – 02:57
Get up time	8:34 am (1:27), 7:15 – 11:58
Time in bed (hours)	8 hrs 33 mins (1:38), 4 hrs 58 mins – 10 hrs 35 mins
Total sleep time (hours)	7 hrs 8 mins (1:27), 3 hrs 52 mins – 9 hrs 11 mins
Onset latency (minutes)	10 mins (0:15), 1 min – 50 mins
Sleep efficiency (percent)	83% (4.57), 76% - 88%
Wake after sleep onset (minutes)	57 mins (0:21), 24 mins – 1 hr 45 mins
No. of wake intervals	25.62 (6.97), 12 – 41.86