Sibling sleep—What can it tell us about parental sleep reports in the context of autism?

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Abstract

Sleep problems are common in families raising children with Autism Spectrum Disorder (ASD). Clinicians often depend on parent reports of child sleep but minimal research exists to address the accuracy or biases in these reports. To isolate parent-report accuracy (from differences in sleep behaviors), the sleep of younger siblings were assessed within a two-group design. The present study compared parent diary reports of infant sibling sleep to videosomnography and actigraphy. In the high-risk group, families had at least one child with ASD and a younger sibling ($n = 33$). The low-risk comparison group had no family history of ASD ($n = 42$). We confirmed comparable sleep behaviors between the groups and used paired t tests, two-one-sided-tests (TOST), and Bland-Altman plots to assess parent report accuracy. The parameters of sleep onset, nighttime sleep duration, awakenings, morning rise time, and daytime sleep duration were evaluated. Diary and videosomnography estimates were comparable for nighttime sleep duration, morning rise time, and awakenings for both groups. Diary and actigraph estimates were less comparable for both groups. Daytime sleep duration estimates had the largest discrepancy with both groups reporting (on average) 40 additional minutes of sleep when compared to actigraphy estimates. In the present study, families raising children with ASD were just as accurate as other families when reporting infant sleep behaviors. Our findings have direct clinical implications and support the use of parent nighttime sleep reports.

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Sleep problems are common in families raising children with Autism Spectrum Disorder (ASD). Parents of children with ASD report more sleep problems in themselves (Bourke-Taylor, Pallant, Law, & Howie, 2013) and their children with ASD (Couturier et al., 2005; Goodlin-Jones, Schwichtenberg, Iosif, Tang, Liu, & Anders, 2009a; Schreck & Mulick, 2000). These reports are often taken at 'face value' with few studies assessing their validity or any reporter bias. Clinicians depend on these reports and a clear understanding of their potential biases is crucial to provide families with optimal sleep education and treatment.

The present study utilizes a two group design and compares maternal diary reports of infant sibling sleep in two groups: families raising children with ASD and at least one younger sibling (high-risk group) and families raising children with no known diagnosis and no family history of ASD at time of enrollment (low-risk group). This infant sibling design is common in ASD research and it is well-suited to address questions of reporter accuracy. When assessing the accuracy of reporters, it is best to have both groups reporting on comparable behaviors (Yoder & Symons, 2010). This is why infant siblings were selected for the current study. Comparisons of infant sibling sleep in high- and low-risk groups are more similar than comparisons between children with and without ASD. If we compare reports of children with ASD to reports of children without ASD it is probable that we would have differences for both the reporter and the base behavior (elevated sleep problems are well documented in children with ASD). This study builds on two research areas including: (a) previous diary comparisons in low-stress homes and (b) stress in families raising children with ASD.

Parent-Report Diary Comparisons

Parent-report diaries of child sleep are commonly used in research and clinical practice. However, the research studies supporting diary use in low-stress homes are mixed. Indices of some sleep parameters (i.e., nighttime sleep onset, morning rise time) have reasonable support (Henderson, France, Owens, & Blampied, 2010; Sadeh, 1996, 2004; Tikotzky & Sadeh, 2001) whereas other indicators like night awakenings have only modest support (Asaka & Takada, 2011; Bélanger, Bernier, Paquet, Simard, & Carrier, 2013; Werner, Molinari, Guyer, & Jenni, 2008). For example, Nelson and colleagues (2014) study of 217 children reported that parents of typically developing children report (on average) 24 minutes more nighttime sleep duration when compared to actigraphy estimates. Conversely, Iwasaki et al. (2010) report parent diary and actigraphy nighttime sleep duration correlations as high as $r = .94$, $p < .001$. For sleep onset time, previous studies document distinct differences between diary and actigraph estimates (Dayyat, Spruyt, Molfese, & Gozal, 2011). For example, Dayyat et al. (2011) compared diary and actigrahy for 327 children and reported diary estimates were (on average) 30 minutes earlier than actigraphy estimates. When considering daytime sleep duration, to our knowledge, no study to date has assessed

the accuracy of these reports. Filling this gap in the literature, we assessed both daytime and nighttime sleep estimates.

When considering parent-reported night awakenings several factors are implicated, including: parental proximity to the child at night, child signaling patterns upon waking, and maternal sleep-related cognitions (Sadeh, Tikotzky, & Scher, 2010; Sadeh, Flint-Ofir, Tirosh, & Tikotzky, 2007; Warren et al., 2003). Specifically, parents who sleep physically closer to their young child are more likely to notice and endorse night awakenings. Similarly, children who signal upon waking are more likely to rouse a parent. It is likely that several factors are reflected in parent reports of night awakenings and should be considered when interpreting them. Studies that compare parent-report diaries to actigraphy indicate that parent reports likely reflect an underestimate of night awakenings (Bélanger et al., 2013; Werner et al., 2008). However, when actigraphy is compared to the field gold standard of polysomnography, actigraphy appears to overestimate night awakenings as it reports extended movements as low-level arousals (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). In the present study, we do not compare actigraphy-indexed arousals to parent reports of night awakenings because it is probable that they index qualitatively different elements of sleep. Parents do not report low-level sleep arousals and actigraphy cannot distinguish between low-level arousals and short awakenings (Sitnick, Goodlin-Jones, & Anders, 2008).

Parent-reported night awakenings reflect the parents' subjective experience of their child's night awakenings; when considering the broader family impacts of child sleep problems this measure may be meaningful for both parental and child health. A recent study by Bourke-Taylor and colleagues (2013) illustrates this. They assessed maternal sleep disruptions and how those disruptions impact maternal well-being/health. In their study of 152 mothers of children with developmental disabilities (94 of whom were diagnosed with ASD), mothers who reported disrupted sleep once per night for four or more nights per week also reported lower levels of well-being/health. Building on this, the present study assesses parent perceptions of child night awakenings and compares them to videosomnography-coded awakenings.

Stress in Families Raising Children with ASD

When working with families raising children with ASD, it is important to assess factors that influence maternal or family well-being/health. Previous research consistently reports elevated maternal strain and lower well-being in mothers of children with ASD (e.g., Duarte, Bordin, Yazigi, & Mooney, 2005). However, previous studies have not indexed how this added stress may impact parental reports of child sleep. In other domains, several studies report associations between elevated parental stress and more parent-reported child behavior problems and restricted and repetitive behaviors (Corcoran, Berry, & Hill, 2015; Lecavalier, Leone, & Wiltz, 2006; Pozo & Sarria, 2015). With respect to sleep, families raising children with ASD consistently endorse more problems (Bourke-Taylor et al., 2013; Couturier et al., 2005; Goodlin-Jones et al., 2009a; Schreck & Mulick, 2000). However, a few studies suggest that subjective parental perception of sleep problems do not align with parental reports of specific sleep behaviors (Goodlin-Jones et al., 2009a; Goodlin-Jones, Tang, Liu, & Anders, 2009b; Schreck & Mulick, 2000). In a study of children with ASD, other (non-

ASD) developmental disabilities (DD), and no known diagnosis, Goodlin-Jones and colleagues reported elevated sleep problems via subjective parent report in both the DD and ASD groups but only found elevated behaviorally-defined sleep problems (e.g., frequent night awakenings) in the DD group (Goodlin-Jones et al., 2009a, 2009b). Similarly, Schreck and Mulick (2000) found that parents of children with ASD reported comparable amounts of sleep in their children when compared to controls but endorsed higher rates of sleep problems. One proposed explanation for this discrepancy is that heightened parental stress levels may contribute to a reporting bias, in which families subjectively report their child's sleep as more problematic than is reflected by behaviorally-defined sleep problems. Collectively, these studies highlight the need to assess parental perceptions of child sleep behaviors, particularly in families raising children with ASD.

In the present study, we do not assess maternal stress or depression but this study is built on the assumption that families raising children with ASD experience, at a group level, higher rates of stress and depression. This position is well-supported by several previous studies (Baker-Ericzén, Brookman-Frazee, & Stahmer, 2005; Bitsika & Sharpley, 2004; Carter et al., 2009; Duarte et al., 2005; Hastings & Brown, 2002; Olsson & Hwang, 2001; Pisula, 2007; Pozo & Sarria, 2015). Additionally, this study assumes this added stress and depression can influence parent perceptions and/or reports of their child's sleep. Previous studies also support this notion with numerous studies documenting differences in parental perception/reports when they are stressed or depressed (Fergusson, Lynskey, & Horwood, 1993; Lee & Hans, 2015; Rutherford, Graber, & Mayer, 2015; Webster-Stratton, 1990). Building on these assumptions, we hypothesized less agreement (i.e., less accuracy) between maternal-report diaries and actigraphy and videosomnography in the high-risk group, when compared to the low-risk group.

To keep our focus on parental sleep reporting and not on differences in sleep behaviors, the present study assessed their younger siblings. This allows for a comparison between reporters (i.e., parents from low- versus high-risk families), without introducing the confound of differences in sleep behaviors across groups. The decision to focus on younger siblings is also a functional one because families were recruited from a larger infant sibling development study.

Method

Participants

As a part of a larger longitudinal study, families were invited to enroll in a supplemental sleep study when their infant was 24 or 36 months of age. Infant age was a function of predetermined developmental check-ups in the larger study and the infant's time of enrollment. Eligible families lived within 1 hour of the University of California, Davis medical center and had no know ASD diagnosis at time of enrollment ($n = 80$). Of these families 75 agreed to participate in the sleep study. The groups included families raising at least one child with ASD and a younger sibling (high-risk group, $n = 33$) and families raising at least two children with no known diagnosis (low-risk group, $n = 42$). In both groups, sleep estimates were collected on the younger sibling. Families were not asked to enroll if the younger sibling had a documented ASD diagnosis to avoid burdening families.

Given the timeframe of this study (an age range that commonly coincides with ASD diagnoses), five children (15%) in the high-risk group received an ASD diagnosis after their enrollment. Given this small sample, we did not do separate analyses on just these children. Additional sample demographic information stratified by risk group is provided in Table 1.

Measures

Family Demographic Information—Infant sex, race, ethnicity, number of children in the home, and family sociodemographic (SES) assets were reported at time of enrollment. Family SES assets included maternal education, paternal education, and family income. Each of these assets were coded (e.g., education was coded: 1 = some high school education, 2 = high school diploma/GED, 3 = some college, 4 = college degree, 5 = some graduate education, 6 = graduate degree). All three were summed with higher scores indicating more assets. Total scores were normally distributed and ranged from 7 to 18 ($M = 12.51$, $SD = 2.75$). Use of composite SES assets scores like these are common in child development research (e.g., Poehlmann-Tynan et al., 2014), particularly when SES assets are not the primary focus of the study but some consideration is desired. Three families did not provide information on family income and one family did not provide paternal education. For these families, the missing values were replaced with the group mean on that variable. Then their SES asset total score was calculated.

Parent-report diaries—Parents were asked to document their infant's sleep behaviors for seven consecutive 24-hour periods using a diary. The diary used in this study is comparable to ones used in previous sleep studies (e.g., Goodlin-Jones et al., 2009a; Schwichtenberg, Anders, Vollbrecht, & Poehlmann, 2011) and included a visual timeline of each 24-hour period and columns to shade-in for infant sleep, sensor removals, sleep in moving objects, and other notes. In this sample, parent informants were predominantly mothers; however, either parent could have completed any portion of the sleep log throughout the week. Additionally, during the day childcare providers could provide information. The diary asked parents/caregivers to document the time their infant was placed in bed at night, sleep onset time, number of night awakenings, morning rise time, and the time their infant left their bed in the morning. Parents/daytime care providers also reported any napping. Data on daycare/childcare use was not collected for daytime sleep duration; therefore, daytime sleep duration reports may reflect naps while in daycare/childcare. Parents were given opportunities to describe any unusual behavior or circumstances (e.g., illness) to determine the typicality of the infant's sleep that day/night. If the infant was reported to be sick during the sleep recording period or if more than 20% of the data were missing, the data were not included in analyses. The descriptive sleep estimates provided in Tables 2 and 3 were generated from diaries with 3 to 7 valid days/nights ($M = 4.71$, $SD = 1.50$). Of the 75 enrolled families only one family did not complete the diary and therefore 74 diaries were available for analyses. Three weekday nights of data was considered the smallest acceptable sleep recording sample for two reasons. First, it allowed us to use data from all of the returned diaries. Second, it allowed us to align diary data with the videosomnography data (which was limited because of the recording storage limitations of the device).

Videosomnography—Videosomnography is an objective measure of sleep that utilizes video recordings to determine sleep-wake behaviors (Anders & Sostek, 1976; Ipsiroglu et al., 2015; Sadeh, 2015). A portable, night-vision camera was placed over the infant's primary sleep location. Videos were coded for sleep onset, morning rise time, and the number and duration of night awakenings. Awakenings had to last longer than 1 minute and had to include purposeful actions from the infant (e.g., sitting up, looking around room, crying or calling out for parents). Videosomnography coders received over 40 hours of training. Inter-rater reliability ranged from .91 to 1.0 with all coders exceeding a predetermined .80 threshold.

Parents were asked to video record their infant's sleep for four weekday nights. Analyses were performed on estimates generated from 3 to 4 nights of data ($M = 3.92$ nights, $SD = .28$). This recording length was chosen based on previous videosomnography studies (Sitnick et al., 2008; Honomichl et al., 2002) and because our recording device could only hold 50 hours of video before it needed to be downloaded. Of the 75 enrolled families, 57 successfully completed the videosomnography recordings. For families who did not complete the video recordings, nine families declined because they did not want their infant on video, five families forgot to turn the unit on for several nights or only recorded partial nights, two families are missing data because of equipment failure, one family's data was not codeable because the camera was moved and recorded another part of the room (not the infant's bed), and one family did not want to have the recording equipment in their home because they feared their older child with ASD would harm the equipment.

Actigraphy—Infant movements during sleep were recorded in one minute epochs using a micromini-motionlogger® (Ambulatory Monitoring, Incorporated). Each infant wore the actigraph on their ankle (imbedded in a neoprene band). The actigraph data were interpreted as sleep or awake using the Sadeh algorithm provided in Action-W version 2.7.3. Following established actigraphy scoring guidelines (Acebo & LeBourgeois, 2006), we used the parentreport diary to index (a) time placed in bed, (b) time removed from bed in the morning, (c) any sensor removals, and (d) sleep that took place in moving objects (e.g., a car or swing). These diary-informed elements helped to clarify the usability of the actigraph data but they did not impact the actigraph sleep onset or morning rise time estimates (which were compared to the sleep diary estimates). Estimates were based on at least three 24-hour periods of valid data (Range 3 – 8 24-hour periods; $M = 5.75$, $SD = 1.25$). For the 75 enrolled families, 68 successfully completed the actigraphy recording week. The seven missing recordings reflected one equipment failure, one parent who did not want their infant to wear the actigraph, and five infants were upset by wearing the actigraph (and it was therefore removed).

Procedure

This study was conducted at University of California, Davis under the approval of the Institutional Review Board and all participating families completed the informed consent process. At time of enrollment into the larger study (when the younger siblings were 6 or 9 months of age), families completed a demographic and family health history form. When these siblings were 24 or 36 months of age, eligible families were asked to participate in a

supplemental sleep study which included two home visits and recording their infant's sleep for seven consecutive 24-hour periods. Families were eligible if they lived within a 1 hour driving radius of University of California, Davis Medical Campus. For enrolled families, the first home visit included setting up the videosomnography recording equipment (i.e., a night vision camera over the infant's primary sleep location). Parents were also given a lightweight, neoprene band containing the actigraph and a sleep diary. They were asked to document their infant's sleep on the diary and to have their infant wear the ankle band for seven 24-hour periods. They were asked to turn on the videosomnography equipment for four weekday nights (e.g., Monday night through Thursday night). Parents were instructed to turn the camera on when they started their infant's bedtime routine and to turn it off in the morning after the infant left their bed. At the second home visit, all of the sleep recording equipment was retrieved and parents received a $25 gift card.

Plan of Analysis

To assess for any differential completion effects for each sleep recording method (diary, videosomnography, actigraphy) a series of ANOVAs were completed comparing those that completed the sleep recording and those who did not. This was possible for the videosomnography and actigraphy data. This was not possible for the sleep diary data because only one family did not complete the requested diary.

To assess parent report accuracy three steps were completed. First, sleep estimates for each recording method were compared across the high- and low-risk groups while controlling for infant sex, number of children in the home, and family sociodemographic assets. This step served to demonstrate that parents in each group were reporting on similar sleep behaviors (e.g., parents in the high-risk group were not reporting on children who had 'worse' sleep than those in the low-risk group). Univariate models were chosen over multivariate models to maximize the number of observations that could be used for each comparison (i.e., to avoid list-wise deletion). Second, sleep estimates in common to each sleep method were compared using a two-tier process (Figure 1). For diary and videosomnography - nighttime sleep duration, sleep onset time, morning rise time, and night awakenings were compared. For diary and actigraphy -nighttime sleep duration, sleep onset time, morning rise time, and daytime sleep duration were compared.

Tier 1 – Statistically Significant Difference—To compare sleep recording methods within groups, first a paired *t* test was used to assess if a statistically significant difference existed between two methods (i.e., diary vs. actigraphy, diary vs. videosomnography). If a statistically significant difference emerged we interpreted this difference

Tier 2 – Equivalence—Lack of a statistically significant difference does not indicate equivalence, it only suggests that the differences between the methods are within the range expected by chance; therefore, two approaches were used to demonstrate comparable reports across the two methods. First, we used the two one-sided *t* tests (TOST) technique (van Stralen, Dekker, Zoccali, & Jager, 2012) to assess equivalence between parent-report diaries with videosomnography and actigraphy. The TOST technique allows us to test the upper and lower bounds of difference against the distributions for each sleep variable. The null

hypothesis for equivalence testing assumes that the difference between measures is outside our predetermined cut-offs. Conducting TOST in each direction allowed us to determine the chance of observing an effect as small or smaller than the one observed. If the p-values are small, then we rejected the null hypothesis and assume comparable measurements (Figure 1). When TOSTs are significant in both directions, this suggests that 90% of the differences between the measures are within the specified range, thus implying equivalence. For nighttime sleep duration, sleep onset, morning rise time, and daytime sleep duration our upper and lower bounds tested were ±30 minutes; for number of night awakenings, we used ½ standard deviation of the sample. These cut-offs followed the work of Bélanger and colleagues (2013) who used similar standards and based on what could be a clinically meaningful difference (e.g., 30 minutes). Our final evaluation of measurement equivalence utilized Bland-Altman plots (i.e., the difference between the paired measurements plotted against their mean value) (Bland & Altman, 1986). This approach provides a visual depiction of the sleep method differences. All analyzed data were synchronized with diary data. For example, if an infant had diary data for Tuesday, Wednesday, and Thursday night then that data was compared to the actigraphy data for the same nights for that infant.

To assess between group differences in reporter accuracy (i.e., measurement agreement) and to confirm these differences do not reflect infant sex, number of children in the home, or family sociodemographic assets we ran a series of linear regressions.

Results

All analyses and assumption checks were completed in IBM SPSS Version 22.

Sleep Recording Method - Differential Completion

To assess for any differential completion effects for each sleep recording method a series of ANOVAs were completed comparing those who completed the sleep recording and those who did not. Infant sex, family risk group status (high- or low-risk), family SES assets, and number of children in the home were not significantly different for the families who did and did not complete the videosomnography recordings (all $p > .15$). For the actigraphy data, infant sex, family risk group status, and number of children in the home were not significantly different for the families who did and did not complete the recordings (all $p > .20$). Families with more sociodemographic assets were slightly less likely to complete their actigraphy recording week, $F(1, 69) = 2.81$, $p = .10$.

Between Group Sleep Estimate Comparisons

To confirm high- and low-risk groups had similar (or at least not significantly different) sleep behaviors a series of univariate models were completed with terms for risk group status, infant sex, family risk group status, and number of children in the home. Tables 2 and 3 summarize these between group comparisons. No significant risk group differences (in any of the assessed sleep estimates) were observed. For morning rise time, one of the overall models did indicate a significant difference (for videosomnography estimates) but this reflected a infant sex difference not a risk group difference. Boys woke earlier than girls, F

(4, 45) = 5.70, $p < .05$. On average, boys woke at 6:49AM and girls woke at 7:42AM (Table 4).

Within Group Sleep Method Agreement

To assess the accuracy of parent-report diaries, agreement between parent-report diaries and videosomnography and parent-report diaries and actigraphy were assessed.

Parent-Report Diaries and Videosomnography—For nighttime sleep duration, diary and videosomnography estimates were comparable for both groups as indicated by paired-sample *t*-tests and TOSTs (Table 2). As illustrated in the Bland-Altman plot (Figure 2a) both groups have comparable agreement for nighttime sleep duration. For sleep onset in the low-risk group, there was no significant difference between diary and videosomnography estimates, $t(31) = -0.99$, $p = .33$; however, the TOST approach indicated the estimates from the two methods were not equivalent (Table 2). In the high-risk group, videosomnography and diary estimates were statistically significantly different, $t(19) = -2.15$, $p = .04$; however, this difference was modest with a difference of about 9 minutes (on average) for sleep onset across the two methods (Figure 2b). For both groups, diary and videosomnography morning rise time estimates were comparable (Table 2; Figure 2c). Night awakening estimates generated from diary and videosomnography were also comparable for both groups (Table 2; Figure 2d).

Parent-Report Diaries and Actigraphy—Estimates generated from diary and actigraphy were comparable for nighttime sleep duration in both groups (Table 3; Figure 3a). In both groups, diary and actigraphy sleep onset estimates lacked agreement (low-risk group, $t(35) = -2.49$, $p = .02$; high-risk group, $t(25) = -3.05$, $p < .01$). On average, parents in the low-risk group reported an earlier sleep onset-time by 10 minutes when compared to actigraphy and in the high-risk group, parents reported sleep onset-time 18 minutes earlier than actigraphy, on average (Table 3; Figure 3b). Our analyses indicated lack of agreement between actigraphy and diary in both groups for morning rise time (Table 3). For morning rise time, the average group differences were 7 minutes later for the low-risk group and 8 minutes later for the high-risk group (Figure 3c). In both groups, estimates for daytime sleep duration across parent-report diary and actigraphy were significantly different (high-risk group, $t(23) = 6.20$, $p < .01$; low-risk group, $t(25) = 4.52$, $p < .01$). Parents in both groups reported more daytime sleep duration, with roughly 40 more minutes of sleep when compared to actigraphy estimates (Figure 3d).

Between Group Sleep Method Agreement

Parent-Report Diaries and Videosomnography—Overall, the differences in diary and videosomnography agreement did not reflect infant sex, family sociodemogaphic assets, number of children in the home, or family risk group status for nighttime sleep duration, sleep onset, or night awakenings (Table 2). Our regression assessing mean morning rise time differences with infant sex, family sociodemographic assets, number of children in the home, and risk group status, $F(4, 47) = 1.93$, $p = .12$, revealed that parents in both groups were less accurate at reporting morning rise time for boys, $t(51) = -2.27$, $p = .03$. Family sociodemographic assets, number of children in the home, and risk group status were not

significant predictors in this model. Parents reported boys woke (on average) 16 minutes later (when compared to videosomnography estimates) (Table 4).

Parent-Report Diaries and Actigraphy—Differences in diary and actigraphy agreement did not reflect infant sex, family sociodemographic assets, number of children in the home, or risk group status for nighttime sleep duration, sleep onset, morning rise time, or daytime sleep duration (Table 3).

Discussion

Following an infant sibling design, the present study assessed the accuracy of parent-report diaries within the context of autism while controlling for base differences in sleep behavior. In sum, the present study indicates that families raising children with ASD are just as accurate at reporting their infant's sleep behaviors when compared to families of children with no known diagnosis. Although agreement across measures varied, the level of agreement was comparable for both groups. Overall, diary nighttime sleep duration reports were comparable to videosomnography and actigraphy estimates. Parents in both groups tended to report earlier sleep onset times when compared to videosomnography and actigraphy estimates and slightly later morning rise times. Though statistically significant, differences in morning rise time and sleep onset were small (in most cases less than 10 minutes), and may not be clinically meaningful. Daytime sleep duration estimates had the largest discrepancy with both groups reporting roughly 40 additional minutes of sleep on the diary when compared to actigraphy estimates. These findings have direct clinical implications in pediatric sleep research and support the use of parent nighttime sleep reports, even in potentially high-stress families.

Unexpectedly parent-reported awakenings did not differ from videosomnography coded awakenings. Given prior findings (e.g., Bélanger et al.2013; Werner et al.,2008) as well as our understanding that only some children signal upon awakenings and others do not, we assumed the videosomnography-coded awakenings would (on average) be more prevalent than parent-endorsed awakenings. Parents in the present study may have been more vigilant in recording awakenings knowing that we were recording their infant's sleep in multiple ways. Alternatively, parents may in fact be valid reporters of the frequency of night awakenings in young children. Prior studies that reported poor agreement in night awakenings between measures included older children (e.g., Werner et al., 2008) who may be less likely to signal during a night awakening or had a rather small sample ($n = 12$; Bélanger, et al., 2013). These findings suggest that clinicians may be able to rely on the accuracy of sleep diary estimates of night awakenings in young children, including families raising a child with ASD.

In applying these findings clinicians and researchers must discriminate between statistical significance and clinically meaningful differences. All of the statistically significant differences were relatively minor with the exception of daytime sleep duraiton. These differences reflect mean-level differences and not individual family differences. However, the Bland-Altman plots illustrate a few families were poor reporters of their infant's sleep behaviors (indicated by dots or squares farther from the mean line) with over an hour

difference in estimates across methods. These clinically meaningful differences create a problem. How do clinicians determine if one particular family is one of these poor reporters? The present study cannot address this issue but future studies should consider other influential family factors (e.g., family chaos) that may aid in identifying less accurate reporters.

The present study informs the broader literature in two main ways. First, we did not find support for bias or less accurate reports from families raising children with ASD. Second, we are the first to compare daytime sleep duration estimates across two methods. The larger daytime sleep duration differences could reflect daytime care location (i.e., childcare or home). Parents may be less aware of their infant's daytime sleep duration when it takes place out of their home. Clinicians should use caution in making treatment recommendations based on sleep diary estimates of daytime sleep duration for young children. Obtaining information directly from other caregivers, such as teachers or childcare workers, may enhance the accuracy of daytime sleep duration estimates. Finally, we illustrated that the comparison method used (i.e., actigraphy or videosomnography) impacts the level of agreement. Parent-report diaries aligned more with videosomnography than with actigraphy. This is not particularly surprising given that both diary and videosomnography index observed human behaviors and actigraphy uses sensor derived movements. Comparisons between diary, actigraphy, and videosomnography in one study are not common; therefore, aligning our findings with the larger field is a bit challenging. However, our study does agree with a theme that carries through the work of others – parent night awakening reports agree with videosomnography estimates (Hednerson et al., 2010; Sitnick et al., 2008) more than actigraphy estimates (Asaka & Takada, 2011; Sadeh, 1996; Titotzky & Sadeh, 2001).

When considering infant sex, two findings emerged with the videosomnography data. Parents in both groups were less accurate at reporting morning rise time for boys and boys woke earlier than girls. As illustrated in Table 4, the earlier rise times of boys appeared across each of the sleep recording methods but to varying degrees. Previous studies of children of a similar age do not endorse this sex pattern for morning rise time (Sadeh, Lavie, Scher, Tirosh, & Epstein, 1991; Taylor, Williams, Farmer, & Taylor, 2015). Similarly, studies that compared parent-report diary to other sleep recording methods while considering sex did not a report differences in morning rise time concordance as a function of sex (Belanger et al., 2014; Nelson et al., 2014). Our infant sex findings appear to be distinctly different from previous studies which may reflect our sample attainment (from a large developmental study with no focus on sleep) or simply a random sampling of boys who woke earlier. Additional research (i.e., replication) is advisable before building clinical recommendations based on these differences.

Limitations of the present study include its relatively small sample size which limited the statistical procedures that could be confidently applied to the data. Additionally, our lack of data on other influential family elements limits our interpretation. Future studies can build on these limitations by incorporating indices of maternal stress or well-being, infant sleep location, and by asking daytime care providers to report on daytime sleep duration. In addition, this study focused on one type of family stressor, caring for a child with ASD, and this may not apply to families facing different types of stressors (e.g., poverty). The present

study may also suffer from enrollment bias. Families who were less stressed may have been more likely to enroll than stressed families. Although enrolled families did not differ based on number of children in the home, sociodemographic assets, or infant sex; we have no index of family stress to assess if families who were less stressed enrolled. The age of enrolled infants (24 or 36 months) should also be considered when interpreting our findings. Infants at this age sleep in a number of locations (i.e., crib, bed, sibling bed) and the present study did not assess how sleep location may influence parent-reports. Nonetheless, with the exception of daytime sleep duration, findings from the present study do support the validity of parent-report diaries from families raising children with ASD.

Two primary clinically-relevant implications of our findings are apparent. First, because our findings indicate that parental reports of nighttime sleep behavior are consistent with objective measurements of sleep behavior, use of sleep diaries and parental reports of nighttime sleep behaviors in clinical practice will likely yield accurate and valid data. Second, our findings indicate that families raising children with ASD are no less accurate at reporting on their infant's sleep behaviors than families raising typically developing children, suggesting that the higher levels of sleep problems reported by parents of children with ASD are likely valid and not merely a reflection of biased reporting. Thus, when parents raising children with ASD report concerning sleep patterns in any of their children, such concerns should be considered a legitimate focus of clinical attention that may be separable from the high stress often experienced by these families and not merely a reflection of generalized "over reporting." Ultimately, it is important to increase our knowledge of the accuracy of parent-reported sleep behaviors in young children, given that other methods for collecting such data are expensive, burdensome, and typically less feasible in clinical practice. Our findings indicate that parent reports of infant nighttime sleep behaviors are reliable, and thus may be validly used to guide initial treatment goals and to monitor ongoing treatment progress, even in potentially high-stress families.

Acknowledgments

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Tier 1 – Paired *t* test
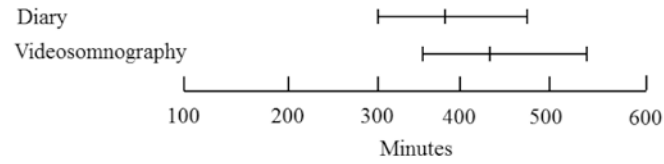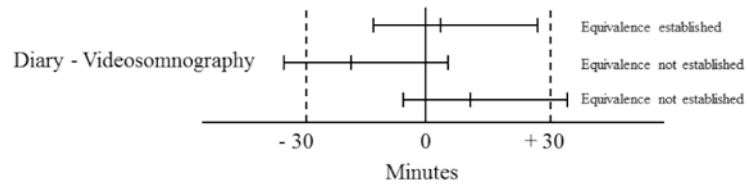This tests if the sleep measurement means (e.g., parent-report dairy and videosomnography) are different from one another (beyond the difference excepted by chance).

Diary
Videosomnography

100   200   300   400   500   600

Minutes

Tier 2 – Two one-sided *t* test (TOST) Approach
These tests assess equivalence by establishing if the difference between the measures falls within the specified range.

Diary - Videosomnography

Equivalence established
Equivalence not established
Equivalence not established

- 30   0   + 30

Minutes

Figure 1.
Conceptual description of the two-tiered process used to compare parent-report diaries to actigraphy and videosomnography.
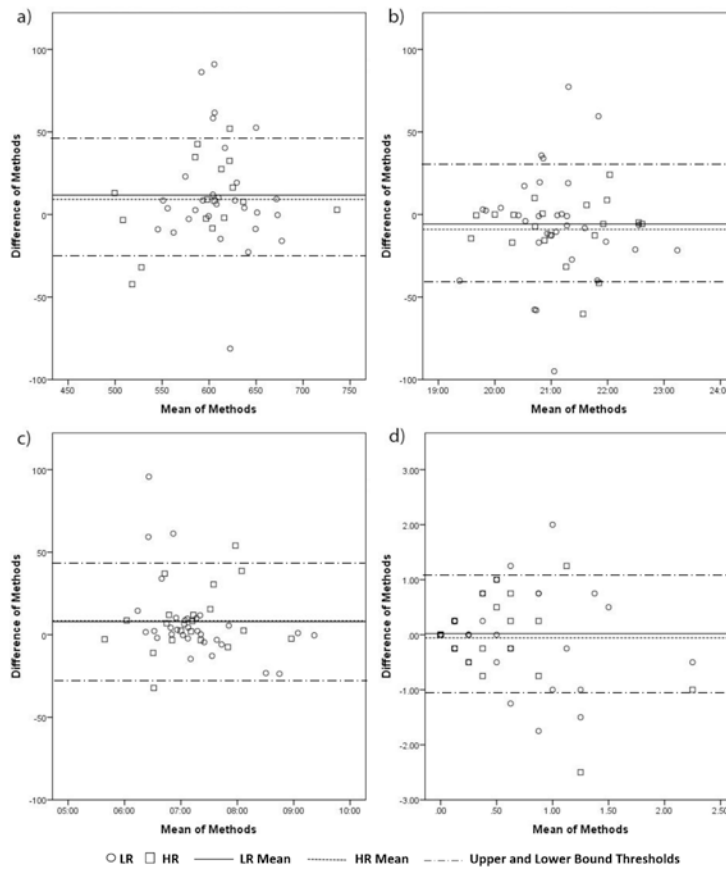
Figure 2.
Bland-Altman plots for diary with videosomnography for low-risk (LR) and high-risk (HR) families for (a) nighttime sleep duration, (b) sleep onset, (c) morning rise time, and (d) night awakenings.
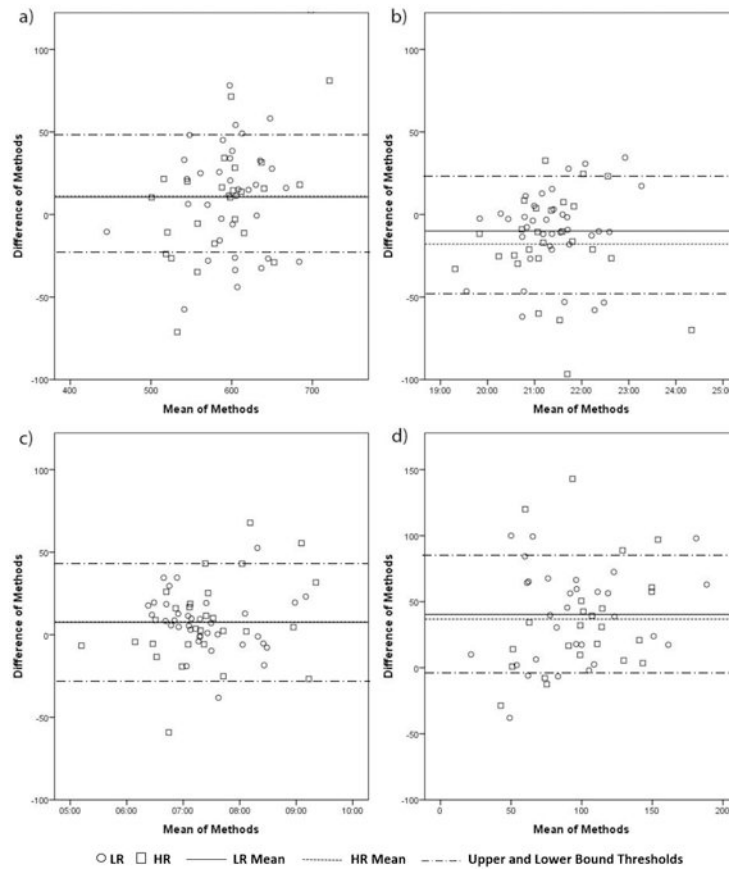
Figure 3.
Bland-Altman plots for diary with actigraphy for low-risk (LR) and high-risk (HR) families
for (a) nighttime sleep duration, (b) sleep onset, (c) morning rise time, and (d) daytime sleep
duration.

Table 1

Demographic Characteristics Stratified by Risk Group (low-risk n = 42, high-risk n = 33)

		Group	
		Low-risk *n*(%)	High-risk *n*(%)
Sex	Male	27 (64%)	21 (64%)
Assessment Age	24 Months	27 (64%)	22 (67%)
	36 Months	15 (36%)	11 (33%)
Race and Ethnicity	Asian	3 (7%)	3 (9%)
	African-American	2 (5%)	0 (0%)
	Caucasian	35 (83%)	27 (82%)
	Multiracial	2 (5%)	3 (9%)
	Hispanic	9 (21%)	7 (21%)
Maternal Education	High School/GED	4 (10%)	2 (6%)
	Some College	3 (7%)	11 (33%)
	College Degree	17 (41%)	14 (47%)
	Some Graduate	3 (7%)	0 (0%)
	Graduate Degree	14 (33%)	6 (18%)
	Other/Unreported	1 (2%)	0 (0%)
Paternal Education	High School/GED	3 (7%)	3 (9%)
	Some College	6 (14%)	8 (24%)
	College Degree	18 (43%)	15 (46%)
	Some Graduate	2 (5%)	0 (0%)
	Graduate Degree	11 (24%)	7 (21%)
	Other/Unreported	2 (5%)	0 (0%)
Family Income	Below $25,000	1 (2%)	2 (6%)
	$25,000-$49,999	3 (7%)	4 (12%)
	$50,000-74,999	12 (29%)	6 (18%)
	$75,000-$99,999	9 (21%)	6 (18%)
	$100,000-$124,999	8 (19%)	5 (15%)
	$125,000 and above	7 (17%)	9 (27%)
	Unreported	2 (5%)	1 (3%)
Number of Children	2	33 (79%)	16 (48%)
	3	7 (17%)	12 (36%)
	4	2 (5%)	3 (9%)
	5	0 (0%)	2 (6%)

Table 2

Diary and Videosomnography Descriptive Statistics and Between and Within Group Comparisons

Sleep Estimate	Nighttime Sleep Duration		Sleep Onset Time		Morning Rise Time		Awakenings	
	HR	LR	HR	LR	HR	LR	HR	LR
	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)
Diary	617(38)	599(62)	21:05 (0:54)	21:01 (0:51)	7:19 (0:41)	7:15 (0:50)	.53(.61)	.48(.56)
Videosom	606(41)	590 (54)	21:14 (0:50)	21:07 (0:50)	7:11 (0:52)	7:07 (0:44)	.51(.69)	.53(.76)
Between Group Sleep Estimate Comparisons [a]								
Diary	$F_{(4, 47)} = 1.87$		$F_{(4, 49)} = .30$		$F_{(4, 50)} = 2.50$		$F_{(4, 66)} = .38$	
Videosom	$F_{(4, 40)} = 1.90$		$F_{(4, 46)} = .59$		$F_{(4, 45)} = 5.70^{*}$		$F_{(4, 50)} = .33$	
Within Group Sleep metod Agreement								
Paired *t* test	$t_{(16)} = 1.54$	$t_{(28)} = 1.85$	$t_{(19)} = -2.15^{*}$	$t_{(31)} = -0.99$	$t_{(19)} = 1.92$	$t_{(31)} = 1.86$	$t_{(21)} = -.34$	$t_{(36)} = 0.17$
↑TOST (+30)[b c]	$t_{(16)} = -3.49^{*}$	$t_{(28)} = -2.88^{*}$	-	$t_{(31)} = -1.08$	$t_{(19)} = 1.81^{*}$	$t_{(31)} = 1.74^{*}$	$t_{(21)} = -2.61^{*}$	$t_{(36)} = -2.99^{*}$
↓TOST (-30)[b c]	$t_{(16)} = 6.58^{*}$	$t_{(28)} = 6.57^{*}$	-	$t_{(31)} = -0.91$	$t_{(19)} = 1.92^{*}$	$t_{(31)} = 1.98^{*}$	$t_{(21)} = 1.93^{*}$	$t_{(36)} = 3.33^{*}$
Between Group sleep method Agreement								
Diary *vs.* Videosom	$F_{(4, 41)} = .42$		$F_{(4, 47)} = 1.06$		$F_{(4, 47)} = 1.93$		$F_{(4, 54)} = .16$	

*
Note. $p < .05$, HR = high-risk group, LR = low-risk group,

[a]
Model with terms for each sleep estimate, risk group, infant sex, number of children in the home, and family sociodemographic assets. TOST = two one-sided *t* tests, ↑ = Upper bound, ↓ Lower bound,

[b]
Upper and lower TOST were only completed if the paired sample *t* test did not indicate a significant difference between the measurement methods. When upper and lower bound TOST are significant it demonstrates that 90% of the difference between the sleep recording methods are within the specified range, thus implying equivalence.,

[c]
for night awakenings .5 SD was used as the upper and lower bounds for the TOST

Table 3

Diary and Actigraphy Descriptive Statistics and Between and Within Group Comparisons

Sleep Estimate	Nighttime Sleep Duration		Sleep Onset Time		Morning Rise Time		Daytime Sleep Duration	
	HR	LR	HR	LR	HR	LR	HR	LR
	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)	M(SD)
Diary	598(72)	602(47)	21:11 (0:59)	21:18 (0:52)	7:28 (1:03)	7:27 (0:42)	120(44)	114(46)
Actigraphy	586(50)	592(46)	21:29 (1:02)	21:28 (0:47)	7:20 (0:54)	7:20 (0:45)	83(34)	74(40)
Between Group Sleep Estimate Comparisons [a]								
Diary	$F_{(4, 61)} = .55$		$F_{(4, 63)} = .67$		$F_{(4, 65)} = .85$		$F_{(4, 61)} = .71$	
Actigraphy	$F_{(4, 56)} = .87$		$F_{(4, 56)} = .65$		$F_{(4, 56)} = 1.89$		$F_{(4, 51)} = .20$	
Within Group Sleep Method Agreement								
Paired t test	$t_{(24)} = 1.39$	$t_{(35)} = 1.99$	$t_{(25)} = -3.05$*	$t_{(35)} = -2.49$*	$t_{(27)} = 1.56$	$t_{(35)} = 2.68$*	$t_{(23)} = 6.20$*	$t_{(29)} = 4.52$*
↑TOST (+30)[b][c]	$t_{(24)} = -2.37$*	$t_{(35)} = -3.69$*	-	-	$t_{(27)} = 1.46$	-	-	-
↓TOST (-30)[b][c]	$t_{(24)} = 5.15$*	$t_{(35)} = 7.67$*	-	-	$t_{(27)} = 1.66$	-	-	-
Between Group Sleep Method Agreement								
Diary vs. Actgraph	$F_{(4, 56)} = .15$		$F_{(4, 57)} = .38$		$F_{(4, 59)} = .45$		$F_{(4, 49)} = .66$	

*
Note. $p < .05$, HR = high-risk group, LR = low-risk group.

[a] Model with terms for each sleep estimate, risk group, infant sex, number of children in the home, and family sociodemographic assets. TOST = two one-sided t tests, ↑ = Upper bound, ↓ = Lower bound,

[b] Upper and lower TOST were only completed if the paired sample t test did not indicate a significant difference between the measurement methods. When upper and lower bound TOST are significant it demonstrates that 90% of the difference between the sleep recording methods are within the specified range, thus implying equivalence..

[c] for night awakenings .5 SD was used as the upper and lower bounds for the TOST

Table 4

Morning rise time estimates used in analyses stratified by sex

Sleep Recording Method	Male	Female
	M(SD)	*M(SD)*
Diary (mornings that align with actigraphy recording mornings)	7:17 (:06)	7:39 (:10)
Diary (mornings that align with videosomnography recording mornings)	7:05 (:05)	7:38 (:10)
Videosomnography	6:49 (:05)	7:42 (:12)
Actigraphy	7:07 (:07)	7:42 (:09)