Symposium Title: Convergent Findings with Different Methodologies when Assessing Sleep in Children with Intellectual and Developmental Disabilities

Chair: Anna J. Esbensen, Ph.D.¹

Discussant: Deborah Fidler, Ph.D.²

Overview: Individuals with intellectual and developmental disabilities (IDD) experience sleep problems at higher rates than the general population (Richdale & Baker, 2014). Risk factors for increased sleep problems are often specific to genetic syndromes (i.e. craniofacial contribution in Down syndrome (DS)) or disorders (i.e. circadian rhythm in autism spectrum disorders (ASD)). In the general population, disrupted sleep is related to daytime behavior, and cognitive and executive functioning (Beebe, 2011). Given the high prevalence of both daily behavior and sleep problems in individuals with IDD, there is growing need to understand the bidirectional relationship between sleep problems and daytime behavior, as well as neurodevelopmental development, in children with IDD. Understanding this relationship is complicated by the multiple methods for assessing sleep problems (polysomnography, actigraphy, sleep diaries). The first presentation examines the impact of sleep problems, as measured by polysomnography, on word learning among preschoolers with DS and among their typically developing peers. The second presentation examines the impact of sleep problems, as measured by actigraphy and parent-report sleep diaries, on challenging behaviors for children with ASD receiving early intensive behavioral intervention. The third presentation examines the impact of sleep problems, as measured by actigraphy and parent report of sleep, on parent- and teacher-reports of maladaptive/adaptive behaviors and executive functioning among school-age children with DS. Using different methods of assessing sleep problems, these presentations demonstrate a convergence of findings, across heterogeneous samples of individuals with IDD, of the impact of sleep problems on daytime behaviors, learning and executive functioning.

References/Citations:

Paper Title: Memory Consolidation across Polysomnography-assessed Naps in Preschoolers with Down Syndrome

Authors: Goffredina Spanò³, Rebecca Gómez², Bianca Demara³, Stephen Cohen³ and Jamie Edgin³

Introduction: Down syndrome is often noted as an intellectual disability impacting memory and learning, including well-documented deficits in hippocampal structure and function (Nadel, 2003). Little previous work has examined processes of sleep-dependent memory consolidation in neurotypical children, and in particular in children with developmental and sleep disorders. At least 50% of young children with DS have impaired sleep, including obstructive sleep apnea (Shott et al., 2006). Previous work has suggested a correlation between sleep disturbance and language learning in this population, including a 9-point difference in Verbal IQ in children with DS and obstructive sleep apnea (OSA) (Breslin et al., 2014) and reductions in vocabulary development in preschoolers (Edgin et al., 2015). DS – characterized by both extensive sleep disturbance and memory impairment – therefore constitutes a good model to examine sleep’s role in memory retention (e.g., memory for new words), allowing us to highlight the processes that may relate to memory formation and retention across both typical and atypical development.

¹ Cincinnati Children’s Hospital Medical Center
² Colorado State University
³ University of Arizona
Methods: We assessed word learning (mapping of object-label associations) in preschoolers with and without DS (25 DS and 25 typically developing (TD) controls) across delays including mid-day naps and wake. Sleep physiology was collected with home-based polysomnography.

Results: Analyses were conducted in 25 children with DS (52% female; mean age = 54.16 mo, SD = 9.49, and range = 41–84 mo) and 25 typically developing (TD) children (52% female; mean age = 33.20 mo, SD = 5.02, and range = 26–50 mo), who completed the three within-subjects study conditions, including memory tests at 5 minutes, after a 4 hour period of wake, and after a 4 hour interval that included a nap (and 24 hours later for each 4 hour condition).

Despite equivalent levels of retention at 5 minutes in the immediate test (t(48) = 1.59, p = 0.12), group differences emerged in the comparison between wake and sleep conditions. We examined group differences on the change in accuracy at the 4-h delay based on the baseline performance at training by performing a 2x2 ANOVA, which revealed a group x condition type interaction, F(1, 48) = 76.26, p < 0.001. No main effects were observed for either the condition type, F(1, 48) = 0.007, p = 0.94, or group, F(1, 48) = 3.36, p = 0.07. Follow-up t-tests showed that children with DS retained less over the nap interval (t(48) = -8.38, p < 0.001; TD > DS), but were significantly better than the TD group over wake (t(48) = 4.66, p < 0.001; DS > TD). While TD performed better after sleep, demonstrating a benefit of the nap (t(24) = -6.72, p < 0.001), children with DS performed significantly better after wake (t(24) = 5.76, p < 0.001). The TD group demonstrated interference across wake, with significantly poorer performance at 4 hours than retention at 5 minutes (t(24) = 2.45, p = 0.02). These results, including the group x condition interaction, persisted at 24 hours.

Discussion: While TD children show interference across a delay containing wakefulness, those with DS show the opposite pattern, and retain more after wake, questioning the value of naps in older children in this group. Despite entering into long intervals of slow wave sleep, children with DS have reduced retention across nap, a finding that adds to our understanding of the dynamics of sleep’s role in memory retention in young children.

References/Citations:
challenging behaviors in this common developmental context for children with ASD. Understanding the reciprocal associations
between sleep and behavior can inform treatment approaches and potentially maximize therapeutic benefits for children with
ASD and their families.

Methods: This study included 40 children with ASD (2-10 years of age). All children attended a center-based intervention
program five days per week. Child sleep was assessed for five consecutive 24-hour periods using an actigraph and a coupled
parent-report sleep diary. During the sleep assessment week, daytime behaviors were recorded during the child’s therapy hours
by his/her behavioral clinician. Daytime challenging behaviors included repetitive behaviors, aggression, negative affect, and
self-injury. Partial–interval recording was implemented in five–minute intervals for the entirety of the treatment day. Clinicians
indicated whether each challenging behavior (e.g., aggression) was observed within each interval. Generalized linear mixed
effects models were used to assess bidirectional relations between sleep and each challenging behavior at both the group
(between-child) and individual (within-child, daily) levels. Sleep parameters, including total sleep time (TST) and wake after sleep
onset (WASO) were included in separate models. Terms for level of adaptive functioning, sex, age, and caregiver education were
also included as covariates.

Results: For between-child effects, children who slept less at night engaged in more repetitive behaviors during the day. On
average during the week, for each hour of missed sleep children engaged in approximately one additional repetitive behavior per
hour ($b = -.96, p < .001$). Additionally, children with higher WASO exhibited more negative affect. On average during the week,
children who woke more at night engaged in .48 more negative affect expressions per hour ($p < .05$). Children who engaged in
more repetitive behaviors and more negative affect during the day also slept less at night. On average during the week, each
missed hour of sleep was associated with 1.5 additional repetitive behaviors per day ($p < .01$). Similarly, on average during the
week, each hour of missed sleep was also associated with 3 additional negative affect expressions per day ($p < .001$). For within-
child effects, children who spent more of their night awake were more likely to engage in self-injurious behavior the following
day. Specifically, for each hour of night waking, children engaged in one additional self-injurious behavior the following day ($p <
.05$).

Discussion: Overall, results from the present study show that sleep and challenging behaviors in children with ASD are
associated. However, in the current study, only WASO and self-injurious behavior exhibited daily associations. This finding
suggests that between-person differences in sleep and challenging behavior mattered more than daily fluctuations in our
sample. Within the treatment context, challenging behaviors (e.g., self-injurious or repetitive behaviors) can influence child
treatment progress and ultimately their developmental trajectory. Findings from the present study highlight sleep as a potential
mechanism to reduce these behaviors. Our findings are consistent with previous research (e.g., Goldman et al., 2011; Mazurek &
Sohl, 2016) and support the use of sleep training or sleep education programs as a part of a child’s early treatment plan.

References/Citations:

  problem behaviors in children and adolescents with autism spectrum disorders. Research in Autism Spectrum Disorders, 5,
  1223–1229. doi:10.1016/j.rasd.2011.01.010
  200. doi:10.1177/10883576050200040101
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Paper Title: Sleep Problems in Children with Down syndrome: The Impact of Raters on Outcomes

Authors: Anna J. Esbensen1 & Emily K. Hoffman1

Introduction: Sleep problems, both OSA and behavioral sleep disturbances are very common in children with DS, often co-exist, and may negatively impact cognitive functioning and daytime behaviors (Esbensen & Schwichtenberg, in press). The impact of sleep on behavior has generally examined young children with DS, and relied on parental report (Stores & Stores, 2004). As sleep problems persist into school-age children, we investigated the impact of sleep, using both parent-report and child actigraphy, on parent- and teacher-reports of daytime behavior and executive functioning among school-age children with DS.

Methods: Participants were 30 children with DS, ages 6-17 years, who participated in a larger study on sleep in children with DS (Esbensen & Hoffman, in press). Children were primarily male (60%), Caucasian (93%) and had a mean age of 11.6 years (SD=2.7). All children wore an actigraph watch at home for a week and parents completed the Children’s Sleep Habits Questionnaire. Parents and teachers completed measures of behavior (NCBRF, CBCL/TRF, Vanderbilt ADHD) and executive functioning (BRIEF). Multiple regression was used to predict behavior and executive functioning from parent-report and actigraphy measures of sleep, controlling for age, gender and race.

Results: Parent ratings of sleep problems were predictive of many parent ratings of behavior problems and executive functioning. Specifically, parent ratings of poor sleep on the CSHQ were predictive of parent ratings of more behavior problems the NCBRF [Calm (β = -52, p < .05), Adapt (β = -52, p < .05), Conduct (β = .56, p < .05), Anxious (β = .61, p < .01), Hyperactive (β = .50, p < .01)], the CBCL [Anxious/Depressed (β = .78, p < .01), Depressed/Withdrawn (β = .56, p < .05), Somatic Complaints (β = .77, p < .01), Thought Problems (β = .60, p < .01), Attention Problems (β = .53, p < .05), Aggression (β = .66, p < .05)], the Vanderbilt [Inattention (β = .58, p < .05), Hyperactivity (β = .50, p < .05)], and of parent ratings of more difficulties with executive functioning on the BRIEF [Inhibit (β = .44, p < .05), Shift (β = .44, p < .05), Emotional Control (β = .45, p < .05), Working Memory (β = .63, p < .01), Organize (β = .48, p < .05)]. In contrast, actigraphy measures of sleep were not predictive of parent ratings of behavior or executive functioning. Rather, actigraphy measures of sleep were predictive of teacher ratings of behavior and executive functioning. Specifically, shorter TST as measured by actigraphy was predictive of teacher ratings of more behavior problems on the NCBRF [Conduct (β = -.66, p < .01), Anxious (β = -.82, p < .01)], the CBCL [Anxious/Depressed (β = -.62, p < .05), Rule Breaking (β = -.55, p < .05), Aggression (β = -.77, p < .01)], the Vanderbilt [Hyperactivity (β = -.55, p < .05)] and of teacher ratings of more difficulties with executive functioning on the BRIEF [Shift (β = -.72, p < .01), Emotional Control (β = -.62, p < .05)]. Sleep efficiency was not related to parent or teacher ratings of behavior or executive functioning.

Discussion: Parents who reported higher sleep problems in their child with DS were also likely to report more difficulties with behavior and executive functioning. However, parent ratings of sleep were generally not associated with behavior reported at school. This finding suggests that shared variance or a bidirectional relationship between behaviors problems and sleep problems is accounting for this pattern. In contrast, actigraphy measures of sleep problems, specifically a shorter TST, were predictive of difficulties with behavior problems and executive functioning at school but not at home. These findings highlight the need to consider multiple raters when measuring outcomes for children with DS, and further highlights the need to consider both parent and actigraphy measures of sleep problems. This information also has a significant impact on our understanding of the role of sleep as a potential confounder in our ongoing measurement of cognitive and executive functioning in children with DS.

References/Citations: