

Title: Correlations between Parent-Reported Sleep and Executive Function in Down Syndrome

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Introduction: Down Syndrome (DS) is one of the most common chromosomal abnormalities in the United States, with 1 in every 700 children diagnosed with DS. The most common deficits seen in DS are congenital heart defects, intellectual disability, gastrointestinal irregularities, hypotonia, decreased visual acuity, thyroid dysfunction, and Obstructive Sleep Apnea (OSA) (Wisniewski et al., 1985). As many as 95% of children have sleep problems as reported by the Child Sleep Habit's Questionnaire (CSHQ), Actigraphy, and polysomnography studies, that may correlate with attention, executive functioning, language development, IQ, memory deficits, emotional control, anxiety, hyperactivity, and daily living skills (Ashworth et al., 2013; Esbensen & Hoffman, 2018; Edgin et al., 2015; Churchill et al., 2015; Chen et al., 2013; Joyce et al., 2020; Esbensen et al., 2017). One area that has been suggested to correlate with sleep is executive functioning. The Behavior Rating Inventory of Executive Function-Preschool and BRIEF Parent for school-aged children questionnaire measure five subcategories for executive function (EF): Inhibit, Shift, Working Memory, Emotional Control, and Planning/Organization. Although studies use both the CSHQ and BRIEF assessments, an analysis between extensive age ranges with a large participant pool is lacking in the literature. This study evaluates the influence of parent-reported sleep problems on adaptive functioning and cognition in DS. It is hypothesized that parent-reported sleep problems will correlate with BRIEF EF measures in children with DS across development.

Method: Actigraphy and BRIEF data from five studies conducted in the Memory Development and Disorders Lab were analyzed. Participants were included in the analysis if they have at least five days of consecutive actigraphy data and completed the age-appropriate assessment. The BRIEF-Preschool was administered to participants up to 6 years old, and the older participants completed the BRIEF-Parent. All analyses were conducted in R.

Results: CSHQ and BRIEF data were available for 112 participants with DS (age range: 2.14-28.13, mean: 10.86), and 138 TD controls (age range: 2.16-17.50, mean: 5.36). 43 participants with DS and 90 TD controls completed the BRIEF-Preschool. 69 participants with DS and 44 TD controls completed the BRIEF-School Age. 48 TD controls completed both questionnaires during different studies. Sleep variables from the CSHQ were significantly different between DS and TD in all domains $F(1, 388-391) = 7.137-63.174, p < 0.05$. Scores on the BRIEF-Preschool were significantly different between both groups, except in Emotional Control, which revealed no significant difference ($w = 2526-3137, p < 0.05$). Similar to the preschool group, DS and TD group differed significantly on all BRIEF-Parent Executive Functioning domains, except for the Emotion Regulation Index and Organization of Materials ($w = 156-2537, p < 0.05$). Sleep-disordered breathing in DS correlated with multiple EF domains in the Preschool group ($r = 0.3721-0.5558, p < 0.05$), but not for the older group. Contrary to expected results, the only domain relating to EF in the older group with DS was bedtime resistance ($r = 0.2861-0.4355, p < 0.05$), while sleep duration, anxiety, and parasomnias correlated with several EF domains in preschool-aged children with DS, ($r = 0.2992-0.4089, p < 0.05$).

Discussion: Similar to previous studies, the results showed a significant difference in parent-reported sleep problems. Overall BRIEF EF scores were significantly different between the DS group and controls in both age groups. In addition, emotional control did not significantly differ in the preschool group between DS and TD but did differ in the school-aged group. CSHQ scores significantly correlated with BRIEF scores in DS and TD in both age groups. However, more CSHQ domains correlated to BRIEF findings in the Preschool group with DS compared to the school-aged group, while the opposite occurred in the TD group. This suggests that poor sleep may correlate with EF differently in DS and TD at different stages of development. Importantly, in DS, sleep-disordered breathing may correlate with EF during the preschool ages, emphasizing the importance for young children with DS to receive sleep studies to screen Obstructive Sleep Apnea. Given these analyses were conducted using parent-report sleep measures known not fully to capture sleep problems in DS, future studies should evaluate further the influence of sleep on EF using both parent-report questionnaires and child-administered assessments of EF.

References:

- Ashworth, A., Hill, C. M., Karmiloff-Smith, A., & Dimitriou, D. (2013). Cross syndrome comparison of sleep problems in children with Down syndrome and Williams syndrome. *Research in developmental disabilities, 34(5)*, 1572-1580.
- Breslin, J., Spanò, G., Bootzin, R., Anand, P., Nadel, L., & Edgin, J. (2014). Obstructive sleep apnea syndrome and cognition in Down syndrome. *Developmental Medicine & Child Neurology, 56(7)*, 657-664.
- Carter, M., McCaughey, E., Annaz, D., & Hill, C. M. (2009). Sleep problems in a Down syndrome population. *Archives of disease in childhood, 94(4)*, 308-310.
- Chen, C. C. J., Spanò, G., & Edgin, J. O. (2013). The impact of sleep disruption on executive function in Down syndrome. *Research in developmental disabilities, 34(6)*, 2033-2039.
- Churchill, S. S., Kieckhefer, G. M., Bjornson, K. F., & Herting, J. R. (2015). Relationship between sleep disturbance and functional outcomes in daily life habits of children with Down syndrome. *Sleep, 38(1)*, 61-71.
- Edgin, J. O., Tooley, U., Demara, B., Nyhuis, C., Anand, P., & Spanò, G. (2015). Sleep disturbance and expressive language development in preschool-age children with Down syndrome. *Child development, 86(6)*, 1984-1998.
- Esbensen, A. J., Hooper, S. R., Fidler, D., Hartley, S. L., Edgin, J., d'Ardhuy, X. L., ... & Rafii, M. (2017). Outcome measures for clinical trials in Down syndrome. *American journal on intellectual and developmental disabilities, 122(3)*, 247-281.
- Esbensen, A. J., & Hoffman, E. K. (2018). Impact of sleep on executive functioning in school-age children with Down syndrome. *Journal of Intellectual Disability Research, 62(6)*, 569-580.
- Joyce, A., Elphick, H., Farquhar, M., Gringras, P., Evans, H., Bucks, R. S., ... & Rush, C. (2020). Obstructive sleep apnoea contributes to executive function impairment in young children with Down syndrome. *Behavioral sleep medicine, 18(5)*, 611-621.
- Wisniewski, K. E., Dalton, A. J., McLachlan, D. C., Wen, G. Y., & Wisniewski, H. M. (1985). Alzheimer's disease in Down's syndrome: clinicopathologic studies. *Neurology, 35(7)*, 957-957.

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