Title: Using Actigraphy to Characterize Sleep in Rett Syndrome

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Introduction: Rett syndrome (RTT) is a neurodevelopmental disorder primarily caused by mutations in the MECP2 gene (Amir, et al., 1999). Sleep dysfunction is commonly reported by caregivers, but findings across studies designed to characterize sleep/wake patterns in RTT tend to be inconsistent and often sleep is measured in different ways (Ellaway et al., 2001). Whether the inconsistencies reflect natural sleep variability as part of the RTT phenotype or measurement differences is not clear. Given the evidence indicating the high prevalence of sleep dysfunction in RTT, there is a potential for clinical trials targeting control of circadian rhythms and sleep/wake cycles. Therefore, there is a need for sensitive, valid, and non-invasive methods of measuring sleep patterns in girls with RTT. As actigraphy has the potential to meet this need, the purpose of the present study was to replicate and extend McArthur and Budden (1998) by using actigraphy to record the sleep patterns of a non-clinical sample of girls with RTT and compare those results to caregiver report.

Method: Thirteen Caucasian females participated in this study (Mean age = 9, 1-17). Caregivers were mailed a Philips Respironics Actigraph 2 device, a sleep diary to record their child’s night and daytime sleep, and questionnaires to gather information about their child’s overall health and behavior. The following variables were derived from actigraphy data using Philips Actiware 6 software: TNS: amount of time spent asleep while in bed; WASO: time spent awake after initial sleep onset; Sleep Efficiency: percent of time spent asleep while in bed; Daytime Sleep: time spent asleep when parents reported a nap. Activity-derived TNS and parent-reported TNS were compared.

Results: Mean total nighttime sleep was 492.3 mins (SD = 47.3) per night, with a mean efficiency of 76.0% (SD = 6.7), and an average of 86.0 minutes (SD = 34.2) spent awake during the night. All participants were reported to daytime sleep during the collection, averaging 46.1 minutes (SD = 50.8). There were no age-related changes for any of the sleep variables measured. Four out of thirteen participants (31%) had mean TNS values that fell within the National Sleep Foundation’s guidelines for recommended TNS. Twelve participants (92%) had a mean sleep efficiency below 85%, a previously used cut off for poor sleep quality (Souders et al., 2009). Parents reported a mean of 97.0 more minutes (SD = 105.2) of TNS and was moderately correlated (ICC = 0.57, p < .001, df = 77), although correlation values differed substantially across participants.

Discussion: This study provides further evidence of the feasibility of using an objective, in-home recording system to characterize sleep in RTT. The results replicated some aspects of previous studies (e.g., no age-related changes in TNS or efficiency). Over half of the sample had total nighttime sleep that fell into the National Sleep Foundation’s recommended range, but most participants had low sleep efficiency. This indicates that some aspects of sleep/wake patterns of girls with RTT may be similar to those of typically developing children, while others may be atypical. Although correlated, parents tended to over report TNS. This indicates that methodology of measuring sleep characteristics may influence results obtained, possibly explaining the inconsistency in the current literature of sleep in RTT. Limitations include a small sample size and a lack of a comparison sample. Future work will add more participants with RTT, as well as an age-matched, typically-developing control sample.

References/Citations: