Title: Stress in Parents of Children with Angelman Syndrome: Considerations of Behavioral, Medical, and Sleep Concerns

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Introduction: Angelman syndrome (AS) occurs as a result of a compromised expression of the UBE3A gene inherited through the maternal side (Bird, 2014). Individuals with AS frequently demonstrate challenging behaviors (e.g., hyperactivity, aggression, self-injury), have significant sleep difficulties, and have medical needs including seizures, all of which result in parent/primary caregiver stress (Goldman et al., 2012; Williams et al., 2006; Wulffaert et al., 2010). Parent reported stress in families of individuals with AS have been observed to be higher than those of other rare disorders and intellectual disabilities (Griffith et al., 2011; Wulffaert et al., 2010). While some research has explored the association between behavioral, medical, and sleep concerns, on parenting stress, limited findings have been described on the cumulative predictive validity of such. The purpose of this study is to explore the relationship between day-to-day concerns experienced by parents/primary caregivers of those with Angelman syndrome and the degree to which these concerns may be predictive of parenting stress.

Method: Families who visited a multi-disciplinary clinic were offered the opportunity to participate in a study aimed at better understanding the clinical needs of individuals and families with AS. Of the 64 individuals and families who consented to have their clinical information used, 48 included information about parent reported behavioral, medical, and sleep concerns with a completed screener of parenting stress. The Parenting Stress Index-Short Form (Abidin, 1995) is a 36-item questionnaire that asks parents to rate the degree to which they agree (strongly agree {1} to strongly disagree {5}) with a statement about their parenting experience, child’s behavior, and distress around experiences. The Behavioral Evaluation of Disorders of Sleep (Shahid, Wilkinson, Marcu & Shapiro, 2011) is a 28-item questionnaire that evaluates sleep problems including sleep disturbances, sensitivity to the environment, disoriented awakening, and apnea. Information on medical and behavioral concerns were obtained from parent report on background information forms.

Results: Scores on the PSI revealed that 25% of the sample experience clinically significant levels of parent distress and 40% had a total stress score considered to be clinically significant. Parent reported behavioral concerns emerged as a positive correlate to parental distress ($r(30) = .41 p \leq .05$), total stress ($r(30) = .47 p \leq .01$), and parent perceptions of having a difficult child ($r(30) = .31 p \leq .01$) where sleep and medical concerns did not. Significant differences between group means of those with and without behavioral concerns were observed in the areas of parent distress ($t(28) = 2.33 p = .03$) and total stress ($t(28) = 2.87 p = .009$), such that parents who reported behavioral concerns had significantly higher levels of parent distress and overall stress. Additionally, behavioral concerns emerged as a significant predictor of overall stress experienced ($R^2 = .22$, $F(1, 28) = 7.86$, $p = .009$).

Discussion: These findings provide further understanding about the rates of parenting stress experienced by parents/primary caregivers of those with AS and how this stress relates to reported behavioral concerns in particular. Future research can be aimed to not only to understand the intersection of behavioral need and parenting stress, but to expand further on what types of behavioral concerns lead to stress including intensity and frequency, and additional conceptualization of these needs to develop targeted interventions for parents.

References/Citations:
