Title: Measuring Early Social Communication Skills in Neurogenetic Syndromes using the CSBS-ITC

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Introduction: Individuals with neurogenetic disorders face a variety of developmental obstacles, including delays in early social communication skills. Identifying appropriate screening measures for monitoring and characterizing infant socio-communicative development in neurogenetic disorders may inform early treatment decisions and improve options for monitoring the effectiveness of clinical trials. The Communication and Symbolic Behavior Scales-Infant Toddler Checklist (CSBS-ITC; Wetherby & Prizant, 2002) is a brief screening measure designed to evaluate risk for communicative and symbolic delays, as well as changes in these skills over time. The measure yields a standardized Total Score and three composite scores: Social, Speech, and Symbolic. The goal of this study was to evaluate the utility of the CSBS-ITC in characterizing development of early social communication skills in infants and toddlers with Angelman Syndrome (AS), Prader-Willi Syndrome (PWS) and Williams Syndrome (WS) by (1) examining whether scores were variable and sensitive to age-related changes and (2) characterizing initial cross-syndrome profiles of early social communication skills in these groups.

Method: Participants included 42 infants and toddlers ages 6-35 months with AS (n=10, $M_{age}=20.1$ months, $SD_{age}=8.37)$, PWS (n=18, $M_{age}=17.56$ months, $SD=7.99)$ and WS (n=14, $M_{age}=22.00$ months, $SD=7.87$). Participants’ mothers were administered the CSBS-ITC as part of an ongoing longitudinal study at Purdue University. Participants >24 months (3 AS, 3 PWS, 6 WS) were included in raw score analyses but excluded from standard score analyses, per publisher guidelines. Analyses were run in R (R Core Team, 2013). We used Spearman correlations to examine whether total and composite scores were sensitive to age, and we used Wilcoxon signed-rank tests to examine pairwise group differences in abilities. Final results will include detailed cross-group and age effects analyses in an expanded sample.

Results: Preliminary results suggest that standard scores cluster around the measure floor in all three syndrome groups, with greatest floor effects for total scores in AS (80% SS 65) followed by WS (38%) and PWS (14%). However as expected, greater variability was observed in raw scores, with scores of zero only observed for the Speech Composite in the AS group (44%). Higher raw scores were associated with older age across Total ($\alpha=.39, p=.01$) Social ($\alpha=.42, p=.01$), and Symbolic ($\alpha=.50, p<.001$) domains, suggesting raw scores are sensitive to age-related development. The correlation between Speech and age approached significance in the whole sample ($\alpha=.26, p=.10$) and reached significance when the AS group was excluded ($\alpha=40, p=.03$), suggesting sensitivity to age in neurogenetic syndromes with more advanced speech. Cross-group analyses suggested similar profiles of early social communication in PWS and WS groups across total and composite standard scores ($p's>.05$). The AS group demonstrated lower social communication skills than PWS, both overall ($W=8, p=.01$) and at the composite level (Social: $W=17, p=.03$; Speech: $W=11.5, p=.01$; Symbolic: $W=5, p=.01$). The AS group also exhibited marginally lower skills than WS ($.06<p<.14$).

Discussion: Results provided preliminary support for using the CSBS-ITC to measure early socio-communicative development in neurogenetic syndromes. Although standard scores exhibited limited variability within each syndrome, raw scores were generally variable and sensitive to age effects, suggesting potential for raw scores to capture developmental growth. The CSBS-ITC was also sensitive to some cross-syndrome differences in our modest sample, warranting further study of whether this measure may be useful in characterizing early development in larger cohorts. Future work should examine the developmental sensitivity of the CSBS-ITC through longitudinal surveillance in expanded neurogenetic samples, as well as by examining its convergence and divergence with other developmental measures.

References/Citations: