Symposium Title: The Severity, Chronicity, and Treatment of Sleep Disturbance in Children With Developmental Disabilities

Chair: Kim Cornish, Monash University
Discussant: Jane Roberts, University of South Carolina

The four presentations in this symposium attempt to address some of the major gaps in our current understanding of the nature and trajectory of sleep disturbance in children with developmental disabilities (DD). The first presentation utilises daily sleep and behavioral data spanning 5 years in over 150 children with Autism Spectrum Disorder (ASD), in order to redefine our understanding of sleep in ASD and provide evidence for distinct sleep profiles. The second and third presentations use actigraphy (a wrist-watch like device which collects movement data every 30 seconds to assess sleep-wake profiles) in conjunction with a typically developing (TD) control group, to quantify the prevalence and severity of sleep in children with ASD. Through a longitudinal follow up, these presentations also allow conclusions to be made regarding the chronicity of sleep disturbance in children with ASD. Finally, in light of the evidence for disturbed sleep as a core feature of developmental disabilities, presentation four summarises the outcomes of sleep interventions for five individuals with a developmental disability and severe problem behavior. Collectively, these presentations reinforce the heterogeneous nature of sleep disturbance in children with developmental disabilities and consequently, the importance of acknowledging the complex and multi-factorial aetiologies of disturbed sleep in this population of children.
**Symposia Title:** The Severity, Chronicity, and Treatment of Sleep Disturbance in Children With Developmental Disabilities

**Chair:** Kim Cornish

**Paper Title:** Characterising Sleep Profiles in 179 Individuals With Low-Functioning Autism Across 100,000 Nights of Sleep

**Author(s):** Simonne Cohen, Monash University  
Kim Cornish, Monash University  
Ben Fulcher, Monash University  
Shantha Rajaratnam, Monash University  
Russell Conduit, RMIT University  
Steven Lockley, Harvard Medical School

**Introduction:** Although there is reason to believe that serious sleep problems are common in children with Autism Spectrum Disorder (ASD)\[1, 2\] and that poor sleep exacerbates challenging daytime behaviour\[3, 4\], such problems have received very little attention in both research and clinical practice. To date, the relationship between sleep and behavioural problems in ASD remains limited to investigations based on small sample sizes, and children with High Functioning Autism. Fortunately, we have access to a large cohort of Low functioning ASD (LFASD) individuals, which will allow us to carefully examine these relationships.

**Methods:** This data has been collected retrospectively from two residential facilities in Boston (USA) which specialize in clinical interventions for individuals with autism ranging from 5-28 years of age. As a part of routine clinical care, sleep-wake behaviour was assessed by continuous clinical observations every 30 minutes from 21:00-7:00h (~110,000 of nightly observations). This is a remarkably comprehensive, high quality dataset that extends for the past 5 years and includes data for more than 179 children.

**Results:** Preliminary analysis of the data found that the average sleep-duration was 9.16±1.28h/night in 5-10 year olds and 8.51±1.49 h/night in 11-17 year olds. 7.83% of sleep-time in the children <11 years, and 50% of sleep-time in the 11-17 year age group did not meet the National Sleep Foundation (NSF) recommended minimum of 10 hours and 8.5 hours of sleep per night, respectively. From this data we can conclude that a proportion of individuals with ASD do not meet the NSF-recommended minimum sleep-duration for age. Cluster analyses of sleep quality indices in 141 LFASD individuals revealed 3 distinct sleep clusters or sleep profiles (Optimal K=3). Cluster 1 (n=35) had a lower mean total sleep time (p<0.001), poorer sleep stability (p<0.001) and sleep efficiency 1-4 hrs into the night (p<0.001) compared to cluster 2 and 3. In contrast, cluster 2 (n=33) had poorer sleep efficiency 5-10hrs into the night (p<0.001) and an early sleep offset (p<0.001) when compared to cluster 1 and 3. Lastly, Cluster 3 (n=74) had higher sleep stability (p=0.05), greater mean total sleep time (p<0.01) and sleep efficiency (p=0.03) compared to cluster 1 and 2.

**Discussion:** Currently, there is an inconsistent understanding of the nature and prevalence of sleep difficulties in individuals with LFASD. These results propose three heterogeneous sleep phenotypes for individuals with LFASD and suggests that poor sleep is prevalent in a large proportion (46%) of individuals with LFASD. We propose that profiling ASD children based on the nature of their sleep disruption will help understand symptom and behavioural profiles (or vice versa) and thus lead to better-targeted interventions. These results will provide the foundation for future randomized clinical trials to test the efficacy of interventional measures to improve sleep and behaviour in children with ASD.

**References:**


**Symposia Title:** The Severity, Chronicity, and Treatment of Sleep Disturbance in Children With Developmental Disabilities

**Chair:** Kim Cornish

**Paper Title:** Profiling the Severity of Sleep Disturbance in Children With Autism Spectrum Disorder: Combined Insight from Actigraphic Monitoring and Parent Report

**Author(s):** Mistral Foster-Owens, Monash University
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Shantha Rajaratnam, Monash University
Kim Cornish, Monash University

**Introduction:** Sleep difficulties are one of the most commonly occurring concurrent clinical disorders in children with Autism Spectrum Disorder (ASD) [1]. The majority of research, examining sleep problems in children with ASD, have done so using parental reports, with the most commonly reported sleep concerns reduced sleep duration due to sleep-onset problems, and poor sleep maintenance [2, 3]. Actigraphy is a relatively non-invasive means to objectively measure the sleep-wake cycle within an individual's home environment. Despite this, to date, only two studies have specifically aimed to use actigraphy to assess sleep-wake differences between school-aged children with ASD and their TD peers [2, 4]. The present study aimed to explore differences in the sleep-wake profile between school-aged children with and without ASD, without ID, using both objective and subjective measures, and explore whether behavioural and environmental factors may be contributing to any sleep-wake differences.

**Methods:** Thirty-four children aged 6-12 years with ASD were compared to 34 age-and gender-matched TD children on objective and parent-rated measures of sleep and behaviour. Parent-reported sleep behaviour was surveyed using the Children's Sleep Habits Questionnaire (CSHQ) and the Bedtime Routines Questionnaire (BRQ). Parents also completed the Strengths and Weakness of ADHD symptoms and Normal behavior scale (SWAN), designed to measure Attention-Deficit Hyperactivity Disorder (ADHD) behaviours. All children completed 14 days of actigraphic monitoring.

**Results:** The prevalence of parent-reported sleep disturbance was significantly higher in the ASD group, with 88 percent of children in the ASD group identified as poor sleepers on the CSHQ (score>41) compared to 44 percent of TD children. Children in the ASD group also demonstrated more parent-reported ADHD symptomology and more maladaptive bedtime routines. Analysis of actigraphic variables revealed that children in the ASD group are put to bed on average 30 minutes earlier than TD children. The ASD group also demonstrated significantly longer sleep-onset delay. While earlier bedtime was significantly associated with more adaptive bedtime routines in the TD group, in the ASD group it was correlated with greater reactivity and more consistent bedtime routine. Shorter weekday sleep-onset delay was significantly correlated with greater parent-reported hyperactivity for the ASD group. In TD children shorter sleep-onset delay was associated with greater parent-reported levels of inattention.

**Discussion:** The current study has shown that parent-reported sleep disturbance is significantly more prevalent in school-age children with ASD, without ID, compared to TD peers. The addition of objective sleep measurement revealed that parents of children with ASD are putting their children to bed earlier than TD children, and sleep initiation difficulties as a prominent feature of the ASD sleep profile. The significant association seen between earlier bedtimes and increased reactivity and consistency in bedtime routine, in children with ASD, likely reflects difficulties commonly experienced by children with ASD, such as such as resistance to change and increased sensory sensitivity, and the subsequent provision of greater settling time by parents. The association seen between greater levels of hyperactivity and reduced weekday sleep-onset difficulties in children with ASD is unexpected and requires further investigation, but may simply reflect increased fatigue due to daytime hyperarousal.

**References:**


Background: Sleep disturbance in children with Autism Spectrum Disorder (ASD) is well established as one of the most prevalent and complex co-morbidities. Particular difficulty in the initiation of sleep is often reported by parents and evidenced by an increased sleep onset latency (SOL)[1]. Few studies have assessed the prevalence and severity of sleep disturbance longitudinally in children with ASD and to date only one study has done so using objective sleep measurement [2]. More longitudinal research is therefore needed to quantify the chronicity of sleep disturbance in children with ASD. Furthermore, it is crucial to capture this trajectory in comparison to that of typically developing (TD) children.

Method: At baseline, participants were 128 children aged 6-12 years, 34 with a diagnosis of Autism Spectrum Disorder and 94 typically developing (TD) children. Parental reported sleep disturbance was assessed via the Children’s Sleep Habits Questionnaire (CSHQ) [3] which was completed by caregivers at two time points, 12-15 months apart. In addition, fourteen nights of actigraphy recording (a small wrist watch like device used to record sleep/wake patterns via movement) were taken at both time points for all ASD participants and a subgroup of TD participants (=75%), to objectively define changes in sleep quality. The longitudinal component of this study (which includes all ASD participants and a subgroup of TD participants) is ongoing and currently includes 54 Participants (21 children with ASD & 23 TD children).

Results: Seventy-one percent of children with ASD had chronic sleep problems as evidenced by scores greater than 41 on the CSHQ across both time points, compared to 30% of TD children. Only 14% of children in the ASD group scored below the CSHQ cut-off at both time points, compared to 52% of TD children. Regarding the initiation of sleep, 62% of children with ASD had chronic difficulties when trying to fall asleep, compared to 22% of TD children.

Within the ASD group, actigraphy revealed a significantly later bedtime and time of sleep onset at follow up, although time of sleep offset and get up time remained stable. As a result, a significant decrease in the amount of time spent in bed (i.e. sleep opportunity) and total sleep duration was observed. Regarding the initiation of sleep, sleep onset issues were chronic over time with an average SOL of around 30 minutes at both time points.

Discussion: As well as reinforcing the increased prevalence of sleep disturbance in children with ASD, these findings highlight the chronicity and severity of such problems, when compared to TD children. Future research should seek to map out this trajectory over a greater period of time, whilst assessing the impact of poor sleep on development. In particular, the direction of this relationship with those psychological (e.g. anxiety) and behavioural (e.g. inattentiveness) factors known to be prevalent in children with ASD, should be further defined.

References:
Introduction: Delayed sleep onset, early awakenings, and waking at night are commonly identified in children with developmental disabilities (DD). Sleep problems may emerge in infancy and early childhood and have been attributed to the rapid growth of the central nervous system (Piazza, Fisher, & Kahng, 1996). Young children with DD are at increased risk for the development of problematic sleep behavior. Irregular sleep patterns have been found to be correlated with the occurrence of severe problem behavior including self-injury, aggression, and disruptive behavior (DeLeon, Fisher, & Marhefka, 2004). Although researchers have examined a variety of sleep interventions with this population, no known studies have looked collectively at multiple cases to explore the relationship between the occurrence of severe problem behavior and sleep patterns in very young children with DD. The current study has two aims: 1) Examine the effect of individualized interventions on targeted sleep behaviors in young children with DD and 2) Assess what influence, if any, sleep interventions had on rates of severe problem behavior prior to the inclusion of additional behavioral interventions.

Methods: Participants included 5 children (2 males, 3 females) referred for assessment and treatment of severe problem behavior. The average age of participants was 2.77 years (Range: 18 months - 4 years). All children presented with a developmental disability. Two children were diagnosed with prenatal exposure to narcotics, two were diagnosed with a genetic syndrome (i.e., Rett Syndrome, Smith-Lemli-Opitz-Syndrome), and one had an autism spectrum disorder. Self-injury was a primary problem behavior for all children; additional problem behaviors included pica (n=2), dangerous acts (n=2) and aggression (n=1). Across the participants, targeted sleep behaviors consisted of night awakenings (n=5), delayed sleep onset (n=4), decreased total hours of sleep (n=4), early awakenings (n=2), and inappropriate location of sleep (n=1). Treatments for sleep problems were individualized across participants; however, all interventions included at least one of the following components: set waking time (n=4), bedtime routine (n=3), response cost with faded bedtime (n=2), no day sleep (n=1), and medication (n=1). Details of the treatment components in addition to the treatment packages for each participant will be discussed.

Results: In all cases, targeted sleep behaviors improved with treatment; in 4 of 5 cases, sleep problems improved with behavioral intervention alone. Graphs of the targeted behaviors pre- and post- treatment will be presented. In addition to targeted sleep behaviors, problem behavior significantly decreased following the sleep treatment in two cases, averting the need for further behavioral intervention. With the remaining three participants, additional behavioral assessment and treatment was required to effectively reduce rates of problem behavior.

Discussion: Findings show that, in certain cases, severe problem behavior can be significantly reduced in young children with DD by regulating sleep. Although problem behavior was not notably affected by a sleep treatment in all participants, results did provide further support for the use of individualized sleep interventions for this population. Implications of findings and directions for future research will be discussed.

References:


Participant ID: 11

Symposium Title: Different Approaches to Better Understanding the Language Skills of Children With Developmental Disabilities

Chair: Lizbeth H. Finestack, University of Minnesota
Discussant: Frances Conners, University of Alabama

The presentations included in the symposium address the need for researchers and clinicians to refine assessments focused on the language skills of children with developmental disabilities. Across the presentations, the language skills of five groups of children are examined, including ASD, FXS, DS, anxiety disorder, and ADHD. Additionally, language is assessed using both standardized language probes and three different language sampling contexts. The areas of language investigated include morphosyntax, narrative language, as well as the use of mazes. Each presenter will discuss the sensitivity of their language measures to differentiate special populations and how performance on the measures adds to what is known about each populations's language phenotype.
Symposia Title: Different Approaches to Better Understanding the Language Skills of Children With Developmental Disabilities

Chair: Lizbeth H. Finestack

Paper Title: A Comparison of the Use of Linguistic Mazes by Children With ASD, ADHD, or Anxiety Disorder

Author(s): Lizbeth H. Finestack, University of Minnesota, Twin Cities
Katherine J. Bangert, University of Minnesota, Twin Cities
Somer L. Bishop, University of California, San Francisco

Introduction: Previous investigators have identified elevated rates of the production of linguistic mazes (filled pauses, repetitions, revisions, and/or abandoned utterances) in the expressive language samples of special populations, including children with autism spectrum disorder (ASD; Sudhalter, Cohen, Silverman, & Walf-Schein, 1990), attention deficit hyperactivity disorder (ADHD; Redmond, 2004), fragile X syndrome (FXS; Sudhalter & Belser, 2001), or conduct disorder (Turkstra, Fuller, Youngstrom, Green, & Kuegeler, 2004). Linguistic mazes are dysfluencies in expressive language that may reflect aspects of executive function such as organization, planning, and memory. Few studies have compared the use of mazes across populations with distinct cognitive and behavioral profiles. Thus, in the current study we compared maze use of children with ASD, children with anxiety disorder, and children with ADHD and examined the relationships between maze use, cognitive profiles, and behavioral profiles in effort to better understand the linguistic and cognitive profiles of these populations.

Method: A total of 48 children aged 5 through 13 years participated in the study. Participation criteria required a diagnosis of one of the following conditions based on the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR; American Psychiatric Association, 2000): ASD (n = 12), anxiety disorder (n = 8), or ADHD (n = 28). Participants were part of a larger study investigating a new diagnostic screening tool for ASD (Bishop, Huerta, & Lord, in preparation). As part of the larger study, all participants completed a full ASD diagnostic assessment, including the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). For the present study, ADOS video recordings were transcribed to obtain child language samples. For each recording, research assistants began transcribing at the beginning of the assessment and stopped transcribing once approximately 100 utterances that contained an overt subject and verb were transcribed. Research assistants transcribed the samples using SALT conventions and researchers used SALT software to obtain key study variable values, including total number of utterances (TNU), mean length of utterance in morphemes (MLUm), type token ratio (TTR), number of mazes per utterance (MAZE), and the rate of mazes per utterance with filler pauses (FP), repetitions (REP), or revisions (REV).

Results: The three groups were well matched based on age (F(2,45) = 0.61, p = .55), TNU produced (F(2,45) = 0.07, p = .93), MLUm (F(2,45) = 0.67, p = .52), and TTR (F(2,45) = 0.31, p = .57), but not nonverbal IQ (F(2,45) = 2.20, p = .12). Thus, nonverbal IQ was included as a covariate in each analysis. Analyses did not reveal significant group differences based on FP (F(2,44) = 0.56, p = .56) or REV (F(2,44) = 0.15, p = .86), but did for REP (F(2,44) = 3.38, p = .04). Post hoc analyses revealed that ASD group produced significantly more mazes with revisions than the children with anxiety disorder and the children with ADHD.

Discussion: Results indicate that children with ASD produce significantly more mazes in their conversational language than other special populations known to have elevated maze rates. However, a significant difference was found only for mazes characterized as repetitions (e.g., (see the) See that cat). These findings may reflect unique executive function profiles associated with ASD.

References:


**Symposia Title:** Different Approaches to Better Understanding the Language Skills of Children With Developmental Disabilities

**Chair:** Lizbeth H. Finestack

**Paper Title:** Narrative Structure in Adolescents With Down Syndrome or Fragile X Syndrome

**Author(s):** Marie Moore Channell, University of California, Davis MIND Institute
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**Introduction:** The phenotypic profiles of individuals with Down syndrome (DS) or fragile X syndrome (FXS) suggest both similarities and differences across the domains of linguistic, cognitive, and social development. Narrative storytelling is an aspect of spoken language that represents the ability to relate a cohesive, sequential story to a listener, a skill that becomes increasingly important to social interactions, especially during adolescence. In general, adolescents with DS or FXS struggle with narrative storytelling. However, very little research has examined whether there are specific aspects of narrative that are especially challenging for individuals with DS or FXS and whether there are differences across the disorders in this regard. Our study examined both macro- and micro-structural components of narratives spoken by adolescents with DS relative to adolescents with FXS as well as younger typically developing (TD) participants matched on nonverbal cognitive ability level.

**Method:** Twenty-three youth with DS (10.28-15.54 years; M = 12.80, SD = 1.59), 22 youth with FXS (10.18-16.01 years; M = 12.33, SD = 1.74), and 23 TD children (3.11-6.19 years; M = 4.48, SD = 0.86) participated in this study. All three groups were matched on nonverbal cognitive ability level (Leiter-R Brief IQ growth scores). Participants completed a Narrative Task in which they viewed a wordless picture book and were instructed to tell the story to an examiner. Audio recordings of participant narratives were transcribed using Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2000) software and segmented into communication units (C-units; defined as an independent clause and its modifiers). Only complete and intelligible C-units were used in the analyses. In terms of macrostructure, we measured story organization by coding each narrative for the presence or absence of story grammar elements important to the sequencing of episodes within the story: Setting, Initiating Event, Internal Response, Plan, Attempt, Outcome, and Reaction. At the microstructural level, we coded the types of words produced (i.e., different types of conjunctions, verbs, and adverbs).

**Results and Discussion:** Preliminary analyses revealed a significant group difference in story organization \(F(2,37) = 3.97, p = .03\). Post-hoc analyses revealed a significant effect of diagnostic group on use of Attempts \(F(2, 37) = 4.01, p = .03\) and Reactions \(F(2, 37) = 3.53, p = .04\), with a marginally significant effect for use of Outcomes \(F(2, 37) = 3.14, p = .06\). Specifically, participants with DS or FXS produced fewer Attempts in their narratives than the TD children, and participants with DS produced fewer Outcomes than the TD children and fewer Reactions than those with FXS or TD. These story grammar elements are necessary for temporal sequencing of events. At the microstructural level, there was a significant group difference in use of conjunctions \(F(2,37) = 5.27, p = .01\) such that the participants with DS or FXS produced proportionately fewer conjunctions than the TD participants. Additionally, there was a marginally significant effect of group on use of verbs \(F(2,37) = 2.89, p = .07\) such that participants with DS produced proportionately fewer verbs than the TD participants.

The preliminary data suggest specificity to the narrative language impairments of individuals with DS or FXS, with different narrative phenotypes across disorders. Future planned analyses include examining the relations between the macro- and micro-structural components of narratives produced within each participant group. We also plan to investigate potential predictors (e.g., age, nonverbal cognition, receptive/expressive vocabulary/syntax) of the between-group differences observed. These data will provide insight into the mechanisms underlying the narrative language impairments observed in DS or FXS, a crucial step toward developing interventions to promote spoken language development and broader social communication skills in individuals.
**Symposia Title:** Different Approaches to Better Understanding the Language Skills of Children With Developmental Disabilities

**Chair:** Lizbeth H. Finestack

**Paper Title:** Methods for Assessing Language in School-Age Boys With Idiopathic Autism and Boys With Fragile X Syndrome

**Author(s):**
Eileen Haebig, University of Wisconsin-Madison Department of Communication Sciences and Disorders
Audra Sterling, University of Wisconsin-Madison Department of Communication Sciences and Disorders
Susen Schroeder, University of Wisconsin-Madison Waisman Center

**Introduction:**
The language phenotype of autism is characterized by delays in expressive language vocabulary, pragmatics, and grammar. This phenotype is closely tied to general language comprehension, and has been compared to other disorders like fragile X syndrome (FXS). Fragile X syndrome is the leading inherited cause of intellectual disability, and there is a significant amount of overlap in terms of the behavioral phenotype between the two disorders. Little research has been done on the best method for assessing language in children with autism with an intellectual disability. Research in fragile X has found that boys perform at nonverbal mental age expectations on standardized tests, but below expectations on a language sample. Language samples require social engagement, and can be particularly challenging for individuals with poor pragmatic skills or those with high levels of social anxiety, both found in autism.

**Objectives:**
To examine the best assessment method for language in children with autism who also have an intellectual disability using a variety of methods commonly used in both the research and clinical domains. We included a comparison group of boys with fragile X syndrome.

**Method:**
Nineteen boys with autism completed the study, as well as 27 boys with fragile X syndrome between the ages of 9-16 years. Assessments included standardized language assessments: receptive and expressive vocabulary (PPVT and EVT), receptive and expressive grammar (i.e., TEGI, CELF), as well as a nonverbal IQ test (Leiter brief IQ), a language sample, and a sentence imitation task. Additionally, each participant completed the Autism Diagnostic Observation Schedule (ADOS) and parents completed the Autism Diagnostic Interview-Revised (ADI-R). The Childhood Autism Rating Scale was completed post-assessment. The language samples, sentence imitation task, and ADOS were transcribed using standard language transcription procedures, and analyzed using the Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2000). Transcripts were analyzed for standard language measures including number of utterances, number of words, grammatical complexity (MLU), and measures of dysfluencies (e.g., repetitive speech, incomplete sentences).

**Results:**
Preliminary results indicate the boys with autism show a relative strength on the sentence imitation task (100% accuracy) and standardized assessments, but lower language levels on the conversation sample (mean MLU: 4.34). The boys with FXS demonstrated more impairment on the sentence imitation task (75% accuracy) and in the language sample (FXS MLU: 3.5).

**Conclusions:**
The boys with autism demonstrated a relative strength on both the sentence imitation task as well as the standardized tests. The language samples yielded less complex language compared to what would be expected based on standardized test performance, but yielded more complexity compared to FXS. We will compare the language used in the language samples to the ADOS in order to look at contextual differences. The boys with FXS struggled with the sentence imitation task, but like autism, demonstrated a relative strength on the standardized assessment. While language samples are an important part of both clinical practice and research, preliminary evidence suggests they are not as informative in terms of the true picture of strengths and weaknesses for some aspects of language, in this case grammar.

This work was supported by grants from the NIDCD (R03 DC011616) and from the NICHD P30 HD03352.
Standardized measures of communication do not always yield useful or interpretable data for individuals with ID. Each of three presentations will share results demonstrating new methods to measure communication. Woynaroski and colleagues found that intraphonological vocal complexity scores derived from LENA software were reliable and stable measures of vocal complexity in children with autism. Duffy and colleagues are developing standardized Expressive language sampling procedures to use as clinical outcomes. Data from participants with FXS reflect high test-retest; and construct validity in comparison to standardized assessments. Brady and colleagues are developing a scale of early communication and report high inter-rater reliability as well as validity in comparison to other measures, based on data from individuals with autism and individuals with ID across a wide age span (3-60 years). Advantages and Disadvantages of the measures will be described as well as plans for further developments.
Symposia Title: New Developments in Measuring Communication

Chair: Nancy Brady

Paper Title: Stability and Validity of Automated Vocal Analysis as a Measure of Vocalization Complexity in Preschoolers With ASD in Early Stages of Language Development

Author(s): Tiffany Woynaroski, Vanderbilt University
D. Kimbrough Oller, University of Memphis
Paul Yoder, Vanderbilt University

Introduction: Explaining individual differences in spoken word use of preschoolers with ASD increases our understanding of variability in this population and helps us predict the extent to which children with ASD will use spoken words to communicate. Theory and research suggest that vocalization complexity explains individual differences in spoken word use in ASD. This longitudinal correlational study examines the relative validity and reliability of four estimates of vocalization complexity in preschoolers with ASD who are preverbal or just beginning to use words to communicate. Three estimates were derived via automated vocal analysis of day-long samples of child vocalizations collected in natural settings, and another was derived from human coded, brief conventional communication samples collected in the lab.

Methods: Participants were 33, 24-48 month old children with Autistic Disorder who were reported by their parents to use < 200 words at entry to the study (Time 1). Our index of vocalization complexity from conventional communication samples was the proportion of communication acts including canonical syllables aggregated with the number of different consonants used communicatively across samples. The first automated index of child vocalization complexity, the infraphonological vocal complexity score, was derived using software developed for research by Oller et al. (2010) that can be applied after standard utterance labeling by LENA software. The additional automated indices of vocalization complexity, the Automated Vocal Analysis (AVA) standard score and developmental age equivalency score, are provided directly by the standard LENA software. Parents reported children's spoken vocabulary use on the MacArthur-Bates Communicative Development Inventories: Words and Gestures (MBCDI) checklist at Time 1 and four months later at Time 2.

Results: The infraphonological vocal complexity score reached our criterion for acceptable stability with one day-long audio recording (g = .82) and covaried with Time 1 (r = .46) and Time 2 (r = .51) spoken vocabulary. Associations for the infraphonological vocal complexity score with concurrent and future spoken vocabulary were non-significantly different from the analogous associations for the variable from conventional communication samples (Z = -1.12, p = .26; Z = -1.02, p = .31, respectively). Vocal complexity measures provided directly by standard LENA software were similarly stable, but were not significantly correlated with concurrent or future spoken vocabulary in our sample. Results were similar for the subset of our sample reported to use < 20 words on the MBCDI at Time 1.

Discussion: Results suggest the infraphonological vocal complexity score from automated vocal analysis is a valid and reliable alternative to the more expensive vocal complexity measures from conventional communication samples and may thus provide a cost-effective method for measurement of vocal complexity in clinical practice. However, at present the infraphonological vocal complexity score is not publicly available.
Introduction: Numerous clinical trials of targeted pharmaceutical agents are now being evaluated for individuals with fragile X syndrome (FXS) and Down syndrome (DS). Disorder-specific behavioral interventions are also being developed. Evaluation of the efficacy of both pharmaceutical and behavioral interventions, however, is hampered by the lack of cognitive and behavioral outcome measures that capture the positive effects of these interventions. In the current study, we present preliminary data aimed at evaluating the adequacy of using naturalistic samples of expressive language to derive clinical endpoints. Expressive language samples (ELS) were collected in two contexts: (a) conversation with an examiner; and (b) narration of a wordless picture book. Such samples reflect important aspects of language competence and atypical language use.

Research Objectives:

1. Examine the test-retest reliability of the ELS procedures.
2. Examine the construct validity of the ELS procedures.
3. Evaluate differences in the psychometric properties of ELS as a function of etiology and task condition (conversation, narration).

Method: Participants: Participants, who were part of a larger study that will eventually enroll 300 participants, had a confirmed diagnosis of FXS (N=11, Mean CA = 12.48; SBS Brief IQ = 49.27, range 47-64) or DS, (N=15; Mean CA = 12.47; SBS Brief IQ = 50.47, range 47-67) ranged in age from 6 - 23 years, were native English speakers, used speech as their primary mode of communication, produced at least some multiword utterances, and had no more than a mild hearing loss.

Procedures: Expressive language samples were collected from participants at two time points, one month apart, by trained examiners blind to participant's performance on standardized measures of language and cognition. Samples were collected in two contexts: conversation with an examiner and narration of a wordless picture book. Examiners followed scripted procedures to ensure comparability of samples across participants. Samples were audio-recorded and transcribed using SALT software.

Measures: ELS dependent measures were computed from SALT transcriptions. Raw scores from standardized language measures were used to evaluate the construct validity of the language sampling variables:

- Syntactic Development -- CELF-4 Formulated Sentences
- Lexical Diversity -- CELF-4 Expressive Vocabulary
- Intelligibility -- GFTA Sounds in Words
- Fluency -- CELF-4 Rapid Automatic Naming
- Talkativeness -- VABS Expressive Language subscale

Results: Data collection and analysis are ongoing, with more participants to be tested prior to the Conference. Here we report preliminary correlations for participants with DS. For conversation, test-retest correlations were significant for intelligibility, lexical diversity, and syntax, ranging from .70 - .95. For narration, test-retest correlations were significant for talkativeness, lexical diversity and syntax, ranging from .70 to .82. Construct validity was high for lexical diversity and syntax in conversation as evidenced by significant correlations with CELF-4 Expressive Vocabulary and Formulated Sentences, respectively. Construct validity was high for intelligibility, syntax, and talkativeness in narration as evidenced by significant correlations with GFTA Sounds in Words, CELF-4 Formulated Sentences, and VABS Expressive Language subscale, respectively.

Discussion: Expressive language sampling has several advantages compared to standardized language tests. Performance on expressive language sampling yields data that is more reflective of performance in functional and meaningful real-world contexts for individuals with intellectual disabilities. Numerous dependent measures, each reflecting a different aspect of spoken language, can be computed from a single language sample, making the procedure ideal for clinical trials, which may impact only some domains of language skill. Expressive language sampling has limited floor effects for individuals producing at least some multiword utterances. The current preliminary findings suggest that expressive language sampling may yield outcome measures ideal for studies of treatment efficacy in individuals with ID.
Introduction: The CCS was created to address a need to measure early communication, particularly in individuals with severe intellectual disabilities. It is a scale that encompasses a range of behaviors from early alerting responses through two word (or symbol) utterances. Previous research indicated that the CCS was reliable and valid with a relatively small sample of participants (Brady et al., 2012). Currently, our team of investigators in Kansas is administering an interactive assessment to 300 individuals between the ages of 3-60 who have minimal verbal skills. In addition, Early Social Communication Scale (ESCS) assessments from young children with autism collected at UCLA, and ESCS assessments from typically developing infants were coded with the CCS. The purpose of this presentation is to present preliminary reliability and validity results, and discuss how these results were used to improve the CCS, as part of an iterative process.

Methods: Preliminary data from three samples were used in the analyses: children with autism from a UCLA intervention study, individuals with a variety of intellectual disabilities from Kansas and typically developing infants (also from Kansas).

Table 1. Demographic information.

<table>
<thead>
<tr>
<th>Sample</th>
<th>Age mean</th>
<th>Age range</th>
<th>% Male</th>
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<tr>
<td>UCLA</td>
<td>3.8 yrs.</td>
<td>3.2-4.5 yrs.</td>
<td>95%</td>
</tr>
<tr>
<td>Kansas ID</td>
<td>23.1 yrs.</td>
<td>4.0 - 60.0 yrs</td>
<td>63%</td>
</tr>
<tr>
<td>Kansas TD infants 1</td>
<td>6 mos.</td>
<td>5.9 - 6.2 mo.</td>
<td>75%</td>
</tr>
<tr>
<td>Kansas TD infants 2</td>
<td>12 mos.</td>
<td>11.9 - 12.1 mo.</td>
<td>57%</td>
</tr>
</tbody>
</table>

We administered the following assessments to each participant in the Kansas sample: Vineland Adaptive Behavior Scale 2, Peabody Picture Vocabulary Test, and the Communication Matrix. We also administered a scripted communication assessment that was later coded with the CCS. 30 assessments were scored a second time for reliability. The UCLA data were coded with typical ESCS scoring conventions as well as the CCS scale. Currently 35 of UCLA ESCS' have been double coded for reliability.

Results: Reliability was calculated in multiple ways including percent agreement, correlation, and Kappa, both for raw scores, and for scores categorized into clinically important ranges (eg pre-intentional, intentional non-symbolic). The range of reliability obtained from the ESCS data indicated that some of the items were not reliable and hence dropped from further coding. Reliability from the Kansas sample was used to identify further training needs including refinement of the original definitions for early communicative behavior. Comparisons of the CCS scores to other measures were used to examine concurrent validity. Preliminary reliability data indicates that for the UCLA data, Kappa for raw scores ranged from .57 to .92. When the broad clinical categories were used, the range was from .66 to complete agreement. Preliminary concurrent validity for the Kansas sample, as indicated by correlations between the average of their highest three communication response scores and existing measures were as follows: Vineland language standard score r=.43**, Communication Matrix average of highest r=.42**, PPVT =-.41*.

Discussion: Preliminary findings were used to inform scoring decisions, and improve scoring criteria and definitions of early communication. For example, disagreements were identified for certain items such as the "creep mouse" activity in the ESCS, leading to a decision to eliminate this item from CCS scoring. Low scoring agreements led to better rules about the time frame for judging triadic eye gaze. Preliminary analyses suggest the CCS will have high concurrent validity with other measures and the scale will be sensitive to developmental differences and intervention outcomes. Our next steps are to complete data collections and analyses regarding validity and sensitivity and begin usability testing aimed at assessing clinical usefulness of the CCS.
A growing body of research provides exciting evidence for the efficacy of both targeted and comprehensive interventions for children with autism, yet relatively few of these interventions are aimed at improving meaningful real-world outcomes for individuals with autism in their naturalistic environments. To a large extent, the degree to which parents, teachers, and other school personnel adopt, implement, and sustain these interventions depends on the fit of these strategies within the context. Conducting research with parameters closer to those seen in real-world practice has the potential that the results will be more relevant, actionable, tailored, and individualized and may contribute to closing some of the gaps between research and practice (Green, 2008; Green, Glasgow, Atkins, & Stange, 2009; Weisz et al., 2004). This symposium will focus on interventions that are situated in homes and schools and mediated through parents, teachers, and other school personnel. The presentations will describe: 1) parent-mediated communication intervention for minimally verbal school aged children; 2) teacher-mediated interventions in preschool and generalized outcomes on core deficits; and 3) school personnel and teacher-mediated social skills intervention at school to improve peer engagement.
**Symposia Title:** Context Matters: Naturalistic Interventions That Address Meaningful Real World Outcomes

**Chair:** Jill Locke

**Paper Title:** Understanding the Mediated Effects of a Speech Generating Device on Social Communication in Minimally Verbal Children With Autism

**Author(s):** Lauren Hampton, Vanderbilt University  
Ann Kaiser, Vanderbilt University  
Connie Kasari, University of California Los Angeles  
Rebecca Landa, Kennedy Krieger Institute  
Danny Almirall, University of Michigan

**Introduction:** Kasari and colleagues examined the effects of naturalistic social communication intervention (JASPER-Enhanced Milieu Teaching; JASP-EMT) delivered with and without an speech generating device (SGD) on the total spoken social communicative utterances (TSCU) for minimally verbal children with autism between 5-8 years old (2014). The intervention included strategies from the Joint Attention, Structured Play, Engagement and Regulation (JASPER) intervention blended with the components of Enhanced Milieu Teaching (EMT) including following the child’s lead, turn taking, building routines, modeling joint attention, using expansions in play and communication, and milieu teaching prompts to teach new language (Kasari et al., 2014). This study analyzes data collected during the randomized trial conducted by Kasari and colleagues (2014). The added benefit of the SGD, or how this addition impacts spoken language, was not readily apparent from the outcome data. A few exploratory studies have found that aided SGD modeling, defined as communication models on an SGD by a communication partner, resulted in improved social communication and comprehension (Drager et al., 2006; Light & Drager, 2007). Thus, we ask the following research question in order to better understand the added benefit of adult communication with an SGD during intervention: Does a change in receptive language mediate the outcome of the SGD intervention compared to the spoken-only intervention on the total use of TSCU?

**Method:** Data for this study were selected from measures collected in the Kasari et al (2014) study at pre-intervention and following 24-weeks of intervention. Three regression models were fit to examine the potential mediation of receptive language on the between groups outcome of increased TSCU. The TSCU was measured in a 20-minute language sample before and after the 24-week intervention. Receptive language was measured by the Peabody Picture Vocabulary Test (PPVT4). The first model confirmed the direct effect of the intervention group assignment (SGD or spoken-only intervention) on TSCU. The second model examined the indirect effect of group assignment on receptive language. The third model examined the effect of group assignment and change in receptive language on TSCU.

**Results:** The regression analyses indicate a significant effect of initial randomization to the SGD intervention on the total social communicative utterances a post-intervention was fully mediated by a change in receptive language (B=0.716, p<.05). The direct effect of intervention group on total social communicative utterances was no longer significant when change in receptive language was added to the model, indicating a fully mediated effect (B=13.623, p>.05).  

**Discussion:** The effect from the primary study, which found that children in the SGD intervention group produced more TSCU than children in the spoken-only intervention group, was fully mediated by the change in receptive language. Thus, the change in social communication for the minimally verbal children with autism was facilitated by the change in receptive language that occurred during the SGD intervention. Implications for practice and future research will be discusses.

**Key references:**


Symposia Title: Context Matters: Naturalistic Interventions That Address Meaningful Real World Outcomes

Chair: Jill Locke

Paper Title: Teacher-Implemented Intervention for Preschool Children With Autism: Engagement and Play

Author(s): Ya-Chih Chang, California State University, Los Angeles
Stephanie Shire, University of California Los Angeles
Wendy Shih, University of California Los Angeles
Connie Kasari, University of California Los Angeles

Introduction: Young children learn through playing; however, children with autism often experience challenges in play. Compared to typically developing children, the play of children with autism can be rigid, repetitive, and tends to consist of lower level functional play acts, lacking symbolic levels of play. Although, there have been many evidence-based interventions focusing on play in preschool children with autism, most of these interventions are conducted in clinics and are done in 1:1 sessions. There has been very limited research examining play in preschool children with autism in a school setting. The aim of this study is to determine if a modified efficacious therapeutic approach (JASPER; Kasari et al, 2010) adjusted for small group instruction is effective in increasing play skills when implemented by teachers in low resourced public preschool programs.

Methods: Seventy-eight preschool children with autism (ages 3-5) were recruited from six autism-specific preschools across the greater Los Angeles area. All preschools included a diverse population of students (91% ethnic minority) and under-resourced students with 80-100% of children receiving free and reduced lunch. A randomized waitlist control design was used. The preschools were matched in pairs based on socioeconomic status and location. Pairs were randomized to either immediate treatment or an 8-week waitlist.

Over the course of 8 weeks, teachers in the treatment condition received 30 x 30-minute JASPER coaching sessions within their existing classroom play rotations with their students. At study entry and exit, blind assessors delivered the Structured Play Assessment (SPA; Ungerer & Sigman, 1981) to each participant. Blind raters then coded play types and level from videotapes. In addition, the first and last JASPER session was taped with each teacher and student. Teachers' (2 at each site) JASPER implementation fidelity was rated from these tapes using a teacher fidelity form to examine seven categories: Basic strategies, Setting up the environment, Following the child's lead, Establishing play routines, Expanding play routines, Responding to communication, and Appropriate use of language.

Results: Generalized linear mixed models (GLMM) with main effects of treatment (IT and WL) and time (entry and exit) were conducted. A treatment by time interaction and subject level random intercepts were used to model the longitudinal trajectories of the outcomes for teacher fidelity and play outcomes (Number of types of Simple, Combination, Presymbolic, and Symbolic play acts). Treatment effects were found for all seven teacher categories (p<.05) and the number of types of Simple (p<.05), Combination (p<.05), and Pre-symbolic (p<.05) play acts. There is a trend toward significance for symbolic play act (p=.1).

Discussion: The results from this study suggest that collaboration with teachers is important, and that teachers can incorporate an evidence-based intervention into their classrooms with proper support in a short amount of time (8 weeks). Most importantly, children with autism are benefitting from the modified intervention that was tailored to their needs including increase in play type and level. Future studies will need to examine how to better support and collaborate with teachers so that evidence-based practices can be better infused and maintained in classroom practices.
Introduction: Although inclusion of children with autism is increasing, inclusion is necessary but not sufficient to improve social functioning (Chamberlain et al., 2007; Kasari et al., 2011). Previous studies have shown that children with autism have a different pattern of inclusion than do typically developing children throughout elementary school (Kasari et al., 2011; Rotheram-Fuller et al., 2010). However, few evidence-based interventions have been successfully implemented in schools because schools often face many challenges when using these programs (Dingfelder & Mandell, 2010). Conducting research in partnership with schools increases the potential that the results will be more relevant to the setting, that schools will actually use the intervention, and that its use will result in positive outcomes for students. Thus, the purpose of this study was to test whether intervention alone or with implementation support improves the social functioning of children with autism.

Methods: Twenty-seven children with autism from grades K-5 and 25 school personnel participated. The average age of children was 8.56 (SD = 2.04) years. The majority was male (89%) and Caucasian (56%). A stepped-wedge randomized controlled design was used where four data points were taken (i.e., baseline, 6-week second baseline, exit, 6-week follow-up). Schools were randomized to: 1) training in Remaking Recess only; or 2) training in Remaking Recess plus implementation support. All school personnel were trained in Remaking Recess during the child’s lunch recess period (approximately 30-45 minutes) for 12 sessions over six weeks. Implementation support was provided to school administrators three times over 12 weeks. Children in the target student’s classroom completed sociometric ratings while blind observers recorded children's solitary and joint engagement with peers on the playground.

Results: Separate multilevel models where time was nested within children and children were nested within schools were used to test differences in social outcomes (i.e. social network salience, solitary engagement, joint engagement with peers, and number of received friendship nominations) between schools that were randomized to Remaking Recess alone or Remaking Recess plus implementation support. There was a significant time by condition interaction where children with autism in schools in the Remaking Recess plus implementation support condition had significantly higher social network salience (F(1, 79) = 5.13, p = .026) and received friendship nominations (F(1, 79) = 4.45, p = .038) compared to the Remaking Recess alone condition. With regard to solitary and joint engagement on the playground, only time was significant (F(1, 79) = 6.93, p = .01 and F(1, 79) = 4.34, p <.001, respectively), where all children with ASD in both conditions spent significantly less time in solitary engagement and more time in joint engagement with peers on the playground.

Discussion: These data provide further evidence that Remaking Recess improves the social engagement of peers on the playground; however, it appears that additional implementation support at the school-level is necessary to improve both social network salience and peer engagement.

Key References:


Children with intellectual disability (ID) and/or autism spectrum disorder (ASD) exhibit hallmark behavioral and cognitive symptoms, which typically serve as the basis for their diagnosis. Other child and family health concerns often take a backseat to the primary features of these intellectual and developmental disabilities (IDDs). However, the mental and physical health of children with IDD and their caregivers must also be considered. The three papers in this symposium consider relationships between child characteristics, disability status, child and parental health, and health care delivery. The first paper examines the trajectories of maternal depression and the transactional relationship between child behavior problems and maternal depressive symptoms longitudinally among families of children with or without ID. The second investigates whether the depressive symptoms of mothers of children with ASD impact the relationship between daily stress and emotional reactivity for these mothers. The final paper explores the trends over time in racial and ethnic disparities in healthcare for children with IDDs. Collectively, we aim to expand the discussion about the true impact of IDD and ASD on health outcomes for children and their caregivers.
Introduction: Few studies in the IDD literature exist tracing maternal depression longitudinally (e.g., Carter et al., 2009; Glidden & Schoolcraft, 2003). Of those published, most are limited to periods either in early childhood or adolescence. Moreover, the extant literature that examines maternal well-being both from cross-sectional and longitudinal perspectives provides some empirical support for reciprocal effects between mothers and their children (e.g., Neece et al., 2012); however, to our knowledge, no one has investigated the transactional relationship between child behavior problems and maternal depression, specifically.

Aims: This study addressed: 1. The change in maternal depression over eight time points and explained the trajectories of mothers’ depression levels utilizing child disability status, child behavior problems, the perceived financial impact of the child, and dispositional optimism as predictors; and 2. The transactional relationship between maternal depression and child behavior problems across the same eight time points.

Methods: Data were collected at child ages 3-9 and 13. Participants included children with ID (N=98) or of typical development (N=141) and their families. Measures included: Center for Epidemiological Studies Depression Scale (CES-D; Radloff, 1977); Child Behavior Checklist (CBCL; Achenbach, 2000; Achenback & Rescorla, 2001); Family Impact Questionnaire (FIQ; Donenberg & Baker, 1993); Life Orientation Test-Revised (LOT-R; Scheier, et al., 1994).

Results: Growth modeling revealed that a linear model was the best fit for the longitudinal data. The final model indicated that child behavior problems had a significant effect on initial levels of maternal depression, predicting above and beyond the child’s age and child’s disability status. Once behavior problems were entered into the model, child disability status was no longer a significant predictor. Inclusion of mothers’ perceived financial impact of the child, after controlling for income, showed that this variable significantly contributed to mothers’ initial depression levels, but not to change in depression over time. Dispositional optimism was found to contribute to both initial depression levels and the change in depression over time. Analysis pertaining to the second aim—investigating the transactional relationship between maternal depression and child behavior problems across time—will include a cross-lagged panel design. This analysis will allow us to determine whether the relationship between these two variables of interest is bi-directional, mother-driven, child-driven, or a function of the stability of the measures alone. Preliminary analysis indicates that both CES-D and CBCL scores are correlated with themselves over time (all p<.001), indicating stability in these variables across the eight time points. Additionally, significant correlations between CES-D scores and CBCL scores at each time point indicate that these variables are significantly related to one another (all p<.001). The cross-lagged panel will assess the longitudinal nature of the relationship between the variables.

Discussion: Both child behavior problems and perceived financial impact of the child were found to predict initial levels of depression in mothers, above and beyond disability status. Only dispositional optimism was found to explain the change in depression over time, indicating that depression trajectories may be better explained by mothers’ dispositional traits, rather than child characteristics.

Key References:


Symposia Title: Health Considerations for Families and Children With IDD

Chair: Anna Esbensen

Paper Title: Depressive Symptoms and Daily Stress Reactivity in Mothers of Children With Autism Spectrum Disorder

Author(s): Naomi V. Ekas, Texas Christian University
Megan Pruitt, Texas Christian University
Lisa Keylon, Texas Christian University
Kelcie Willis, Texas Christian University

Introduction: The ability to regulate emotions has important implications for an individual's overall functioning (physical, mental, and social). Mothers raising a child with autism spectrum disorder (ASD) face unique challenges that may impact their ability to effectively regulate their emotions. These chronic challenges may also translate into elevated levels of stress, depression, and anxiety. Individuals with depression tend to be more emotionally reactive to stressful events on a daily basis (Bylsma et al., 2011). Recent studies have examined the daily experiences of mothers of children with ASD and found that mothers' perceptions of stress predicted negative affect (Ekas & Whitman, 2011). However, there have been no studies examining whether mothers' depressive symptoms affect the associations between daily stressful events and emotional reactivity. The current study examines this research question using daily diaries and includes both positive and negative emotional reactivity.

Method: Forty-nine mothers of a child with ASD participated in the current study. Mothers completed an initial questionnaire assessing their current depressive symptoms (CESD). Mothers then completed 30 consecutive days of daily diaries. Each day mothers answered questions about the emotions they experienced that day (PANAS). In addition, each day mothers completed a checklist of daily hassles where they indicated whether the stressful event had occurred during that day.

Results: Hierarchical linear modeling (HLM) and linear regression analyses were used to test our research questions. In HLM, the level 1 model specified the relationship between daily stress and daily affect. Separate models were tested for negative and positive affect. Results indicated that for each additional stressful event that occurred on a given day, mothers negative affect increased 1.60 points (t(48) = 6.23, p = .00) and positive affect decreased .48 points (t(48) = -2.33, p = .024). We then generated Bayesian-based emotional reactivity slopes to use in subsequent analyses. We used Bayesian slopes as they are considered to be more accurate than OLS slopes. Using linear regression we examined whether depressive symptoms predicted daily emotional reactivity. Results indicated that higher levels of depressive symptoms predicted increased negative reactivity (b = .03, p = .033) and increased positive reactivity (b = .02, p = .00).

Discussion: The results of the current study suggest that mothers of children with ASD are emotionally reactive to stressful events that occur on a daily basis. When examining the impact of depressive symptoms, we found that elevated symptoms predicted heightened emotional reactivity. These results are consistent with studies of adults in the general population. Heightened reactivity to stressful events may spill-over and impact mothers' interactions with family members, including their children. We plan to conduct further analyses to determine whether other maternal characteristics, such as self-esteem or sense of control, predict daily emotional reactivity.

Key References:


**Symposia Title:** Health Considerations for Families and Children With IDD

**Chair:** Anna Esbensen

**Paper Title:** Trends in Racial/Ethnic Disparities in Quality of Provider Interactions for Children With Autism and Developmental Disabilities

**Author(s):** Sandy Magaña, University of Illinois at Chicago  
Esther Son, College of Staten Island, CUNY  
Susan Parish, Brandeis University

**Introduction:** The American Academy of Pediatrics (2012) released a policy statement highlighting the importance of family-centered care across all pediatric settings and high quality provider interactions are core features of family-centered care. Recent research has found significantly reduced health care access, lower service utilization, and worse quality in provider interactions among African American and Latino children with autism and developmental disabilities (DD) compared to White children, even after controlling for important confounders including parent education, income, and severity of the child's condition. However, researchers have not investigated population trends for these racial and ethnic disparities. Using data from the 2005/06 and 2009/10 National Survey of Children with Special Health Care Needs (NSCSHCN), we examined changes in disparities in the quality of provider interactions for African American and Latino children with autism/DD compared to White children with autism/DD. Our research questions are: 1) Are there disparities in the quality of provider interactions for African American and Latino children with autism/DD in 2005/06 and 2009/10 NSCSHCN?; and 2) Did the racial/ethnic disparities in the quality of provider interaction change between the two years?

**Method:** Our sample consisted of children with autism/DD (n=12,172) from NSCSHCN. Descriptive and multivariate logistic regression analyses were conducted using Stata 12.0 to account for the complex sampling design. A variable for survey year was used to determine whether there were statistical differences between 2005/06 and 2009/10 after adjusting for all model covariates (child's age, gender, and severity of condition, family structure, parental education, household income, lives in a rural/urban area). An interaction term (year × ethnicity) was modeled to identify statistical difference across years and ethnic groups.

**Results:** After controlling for all covariates, we found that racial and ethnic disparities in the quality of provider interaction were substantial in both 2005/2006 and 2009/2010 years. Black and Latino parents were significantly less likely than White parents to report that their provider spent enough time with child and was sensitive to family's values and customs. Racial and ethnic disparities in the quality of provider interactions were found to be unchanged over time.

**Conclusion:** This study found new evidence that racial and ethnic disparities in the quality of healthcare provider interactions with parents of children with autism/DD persist. Despite ongoing calls for interventions to reduce such disparities; there is no evidence that these disparities are being reduced over time. The need for policy makers to implement aggressive measures to remedy this situation is imperative.
Beginning as early as the preschool years, children with autism or intellectual disabilities enter school on an uneven playing field with differing formal educational experiences (Burgess, Hecht, & Lonigan, 2002). Children with autism demonstrate deficits in social skills, and often struggle to connect with teachers and learn from teacher-child interactions (Pianta & Stuhlman, 2004). Varying skills and deficits in behavioral, academic, social and communication outcomes also impact children with developmental delays and autism (Nation, Clarke, Wright, & Williams, 2006). With a growing push to level the playing field, children's school preparedness and positive relationships with parents and teachers have become areas of interest, and may impact both academic and nonacademic outcomes (Anderman, 2002). The three studies in this symposium examine disparities among children with intellectual disability (ID) or autism spectrum disorders (ASD). The first study examines strategies that parents use to develop early language and reading skills in young children with ASD, despite the impact of social or behavioral challenges. The second study examines the relationship of classroom and school placement decisions upon school entry to child social development and academic growth over time. The third study then extends these experiences to adolescence, and identifies variables related to school climate and behavioral characteristics of youth with and without ID or ASD. Together, these studies help elucidate the broader implications of delays in children with ID and ASD, as well as the strategies and support parents and schools provide to close the gap in adjustment over the course of development.
**Symposia Title:** School Daze: Autism, ID, and Educational Issues

**Chair:** Laura Lee McIntyre

**Paper Title:** Shared Book Reading and the Child with ASD: The Parent's Role

**Author(s):** Leigh Ann Tipton, University of California-Riverside  
Jan Blacher, University of California-Riverside  
Abbey Eisenhower, University of Massachusetts-Boston

**Introduction:** Shared book reading allows parents to structure reading activities to help children achieve higher level skills for reading. Whitehurst and colleagues (1988) conducted an early systematic study that examined the effects of shared book reading on the development of language (and ultimately reading) in young children. They found that parents were able to elicit higher level skills in their children (e.g., by using open-ended questions) and to increase praise and feedback contingent upon the child’s communication attempts.

The purpose of this study was to examine parent and child behaviors during a shared book reading activity with young children on the autism spectrum. The focus was on the following questions: (1) What types of language or literacy-related behaviors do parents of children with ASD provide during a shared reading task? When coded, do these behaviors replicate a two-factor model of language elicitation techniques (evocative and feedback strategies) previously used with typically developing children in a similar context? (2) To what extent do spoken language, behavior problems, and social skills of children with ASD relate to parents’ shared literacy task strategies? (3) To what extent do parent shared literacy reading strategies relate to child early academic skills?

**Method:** The current study included families from a longitudinal study of early transition to school for children with autism spectrum disorders (ASD). The sample (N=111) included children who had (a) diagnosis of autism, confirmed with the ADOS and/or ADI-R, (b) chronological age between 4 and 7 years 3 months, (d) functional verbal ability of three words or more, and (e) intelligence quotient (IQ) greater than or equal to 55. Parents participated in a shared-book reading activity. The system designed by Whitehurst et al. (1988) was used to code observed parent behaviors, facilitative of language and reading development, during shared book reading.

**Results:** Parents' demonstrated high use of reading/conversation throughout the task (100%) but relatively few attempts to provide imitative directions (27.9%; e.g., is he happy or surprised?) The results of a factor analysis identified a four-factor model of a parent-directed behavior during a shared literacy task—clarification techniques, feedback techniques, teaching techniques, and evocative techniques. These four factors explained 62.8% of the variance of the overall shared literacy model. (2) Child characteristics related to parents' shared book reading strategies as demonstrated by regression analyses; parent education levels and child social interaction skills related to parents' use of clarification techniques during book reading (as also demonstrated that in predicting children's early phonics skills, parent's use of feedback techniques and child IQ status were both significant predictors in a regression model (R2=.15, F=8.45, p<.001).

**Discussion:** Parenting behaviors as demonstrated during shared book reading can support early literacy development in their children with ASD. Surprisingly, characteristics of the child with ASD contributed less to the variance in parents' shared reading behaviors than home literacy practices. Furthermore, the addition of parent’s shared reading techniques, despite any formal intervention, was able to increase children's academic readiness skills.

**Key References:**


**Symposia Title:** School Daze: Autism, ID, and Educational Issues

**Chair:** Laura Lee McIntyre

**Paper Title:** Classroom Placement: Implications for Academic Growth and Student Outcomes in Students With ASD

**Author(s):** Geovanna Rodriguez, University of California-Riverside  
Erin Knight, University of California-Riverside  
Abbey Eisenhower, University of Massachusetts-Boston  
Jan Blacher, University of California- Riverside

**Introduction:** Children with autism spectrum disorders (ASDs) exhibit impairments in socio-communicative functioning, social interaction, and restricted, repetitive behavior (5th ed.; DSM-5; American Psychological Association, 2013). However, teachers and parents often perceive co-morbid behavior difficulties as more problematic than core autism features (Pearson, Loveland, & Lachar, et al., 2006), which can affect placement decisions and overall school functioning. Furthermore, deficits associated with ASD are related to academic difficulties (Nation, Clarke, Wright, & Williams, 2006).

Despite research on the benefits of general education settings (Stahmer & Ingersoll, 2004), the early school transition period has not been examined in children with ASD. Research remains largely inconclusive as to what factors determine classroom placement, and the overall role of placement in impacting skill acquisition over time (Kurth & Mastergeorge, 2010). The proposed study aims to examine factors related to placement decisions, and the impact classroom placement has on student outcomes over time. The following questions will be addressed:

1) To what extent do comorbid issues (externalizing/internalizing behaviors) predict child classroom placement above and beyond child ASD characteristics (i.e., IQ, language level, and ASD symptomology)?
2) To what extent does child classroom placement predict child social and academic outcomes over time?

**Methods:** Participants were recruited from a larger study of the early transition to school for children with ASD (N = 103), most with high cognitive functioning in the typical range (M IQ = 90.54), from 4 to 7 years old (M = 5.3 years). Participants were assessed at three time points over two school years (beginning of year 1, end of year 1, and end of year 2). Literacy on the Woodcock-Johnson III Tests of Achievement, Passage Comprehension (McGrew, Schrank, & Woodcock, 2007), comorbid issues (externalizing and internalizing behavior problems on the Child Behavior Checklist; Achenbach & Rescorla, 2000; 2001), ASD characteristics on the Social Responsiveness Scale (Constantino & Gruber, 2005), and placement (report of general education or special education) were measured at multiple time points.

**Results:** A logistic regression predicting Time 3 classroom placement (general education or special education) using ASD characteristics as predictors (i.e., language level, IQ, and autistic mannerisms), suggested that only autistic mannerisms (β = .04, p = .08) approached significance, $\chi^2 (3) = 28.77$, $p < .001$. The logistic regression using CBCL internalizing and externalizing t-scores as predictors of placement was not significant, $\chi^2 (2) = 1.60$, $p = .45$.

A hierarchical regression was run to predict Time 2 social skills using Time 1 classroom placement while controlling for Time 1 social skills; although the model was significant, F(2, 95) = 62.93, $p < .001$, classroom placement was not a significant predictor, $\beta = .01$, $p = .92$. A hierarchical regression predicting Time 3 reading comprehension using Time 1 classroom placement while controlling for Time 2 reading comprehension was significant, F(2, 100) = 45.38, $p < .001$, and classroom placement predicted comprehension above previous comprehension, $\beta = -.18$, $p < .05$. Latent growth curve analyses will be run to examine this more thoroughly.

**Discussion:** Although preliminary analyses suggested that comorbidities do not impact placement decisions, academic growth may be affected by child placement. Thus, it is important for parents and schools to consider the consequential validity of school placements for children with ASD.

**Key References:**


Introduction: Recently, there has been growing interest in the effects of schooling on nonacademic outcomes for children such as mental health and behavior problems (Anderman, 2002). One longitudinal study found that positive perceptions of school social climate moderated negative effects of students’ self-criticism on internalizing and externalizing problems (Kuperminc, Leadbeater, & Blatt, 2001). The significance of school climate (including school connectedness and relationships with teachers and peers) has been studied in typically developing (TD) adolescents, but little research has extended these questions to adolescents with autism spectrum disorder (ASD) and/or intellectual disability (ID). The present study had the following aims: 1. To examine differences in aspects of school climate experienced by each disability group (ASD, ID, and TD); and 2. To determine the relationship of school climate variables to behavior problems and psychopathology for these students.

Methods: Participants included 101 youth: adolescents with ASD (N = 26; mean IQ = 84.8), ID (N = 19; mean IQ = 58.5), and TD (N = 56; mean IQ = 111). Researchers conducted in-depth adolescent interviews about friendships and school experiences when the youth was both 13 and 15 years old. Mothers completed the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001) and the Diagnostic Interview Schedule for Children Version IV (DISC-IV; NIMH); youth completed the Youth Self Report (YSR; Achenbach & Rescorla, 2001) and People In My Life (PIML; Cook et al., 1995). The academic subject teacher closest to the youth completed the Student Teacher Relationship Scale (STRS; Pianta, 2001) and Teachers’ Rating Scale of Social Acceptance (TRS; Harter, 1982).

Results: Regarding aim 1, a 3-way ANOVA found significant group differences by disability status in STRS conflict (F = 5.52, p < .01) and dependency (F = 10.43, p < .001), and in the teacher social acceptance total score (F = 15.8, p < .001). Post-hoc analyses revealed the ASD group had significantly lower bonds with school, higher STRS conflict, and lower teacher rated peer acceptance, than the ID and TD groups; both the ASD and ID groups had significantly higher STRS dependency than the TD group. Regarding aim 2, preliminary analysis indicated significant negative correlations between total behavior problems at age 13 and youth rated affiliation with teacher (r = -.23, p < .05) and bonds with school (r = -.36, p < .01), STRS total score (r = -.29, p < .01), and teacher rating of social acceptance total score (r = -.41, p < .01). There were significant positive relationships between total behavior problems at age 13 and STRS conflict (r = .32, p < .01) and STRS dependency (r = .37, p < .01). Further analyses will include multiple regressions examining the relation of school variables at age 13 to behavior problems and psychopathology for adolescents at age 15.

Discussion: Initial analyses suggest that students with ASD experience a less positive school climate than students with ID or TD. Predictors of outcomes at age 15 will suggest directions for future research and intervention in school climate.

Key References:
Analogous to the study of behavioral phenotypes in which different neuropsychological and behavioral characteristics are associated with different developmental disorders, researchers studying the neuroanatomical phenotypes in these groups have identified neural phenotypes that are shared across different disorders (e.g., vulnerabilities in the superior temporal and dorsolateral prefrontal cortices and in the cerebellum). Understanding the unique and shared behavioral and neural phenotypes associated with developmental disorders may provide insights into etiological overlaps and how to tailor treatments for youth with specific disorders as well as identify aspects of cognition and behavior that may respond to similar treatments (e.g., educational, behavioral, and/or biomedical) across groups. Thus, in the current symposium, we seek to examine cortical and cerebellar phenotypes associated with five developmental disorders - autism spectrum disorder, Down syndrome, 49, XXXXY syndrome, dyslexia and attention-deficit/hyperactivity disorder. While each of these disorders constitutes a separate diagnostic category (with some disorders diagnosed based on phenotype, others on genotype), they share features at the neuropsychological and neuroanatomical level that may be helpful in determining etiological similarities and identifying treatment targets for cognitive and behavioral constructs that cross diagnostic categories (e.g., executive dysfunction and/or language-based learning difficulties).
Symposia Title: Using Structural MRI to Identify Shared and Unique Neuroanatomic Vulnerabilities in Five Developmental Disorders

Chair: Nancy Raitano Lee

Paper Title: Shared and Unique Characteristics of Cortical and Cerebellar Anatomy in Down Syndrome and 49,XXXXY Syndrome: A Pediatric Neuroimaging Investigation

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Introduction: Down syndrome (DS) and 49,XXXXY syndrome are two chromosomal aneuploidies associated with reduced cortical volumes [1,2]. However, most existing studies have described these features at the level of gross brain anatomy (e.g., lobar-level descriptions). In an effort to refine descriptions of the brain in young people with these syndromes, the current study had the following aims: (1) provide descriptions of cortical thickness (CT) and surface area (CSA) measured at the global and vertex-level (at ~81,0000 vertices across the cortex) in both groups; (2) contrast regional patterns of CT and CSA overlap and divergence in DS and 49,XXXXY; (3) begin to characterize the cerebellum in these groups.

Methods: Participants with DS and 49,XXXXY participated in two studies at the National Institutes of Health. As a result, different magnetic resonance imaging (MRI) scanners were utilized. Thus each group was compared to a sex- and age-matched control group with data acquired on the same scanner. Cross-syndrome comparisons were completed by contrasting z-scores relative to respective control groups for males with DS and males with 49,XXXXY (an exclusively male disorder). Participants included 31 youth with DS and 45 typically developing (TD) age- and sex-matched peers (M Age=15; Range=5-24 years; 34 males) scanned on the same General Electric (GE) 3T scanner. In the 49,XXXXY study, participants included 7 males with the syndrome and 24 age-matched male TD peers (M age=15; range 8-17 years) scanned on the same GE 1.5T scanner. All brain scans were processed with Montreal Neurological Institute’s CIVET pipeline. Group differences were examined using linear regression with the following equation: CT/CSA/Cerebellar Volume ~ Sex (for DS-control comparisons only) + Age + Age squared + Group.

Results: For DS, cortical volume was reduced by 7%. However, a dissociation between CSA and CT was found. CSA was largely reduced while CT was largely increased (p<.001). These findings were observed at the whole cortex and vertex-level. Pronounced areas of CSA reduction were identified in the superior temporal gyrus and frontal lobe. In contrast, CT was largely increased, particularly in anterior and posterior brain regions (FDR-corrected qs <.05). Cerebellar volume was reduced by 26% in DS (p<.001). For 49,XXXXY, cortical volume was reduced by 20%. Unlike DS, both CSA and CT were reduced in 49,XXXXY, though only CSA reached statistical significance at the whole cortex level (p<.001 for CSA; p=.15 for CT). Syndrome comparisons of vertex-level z-scores (relative to controls) revealed regionally specific divergence for CT primarily in anterior and posterior brain regions. Specifically, the DS group had largely increased CT in frontal and parietal-occipital regions compared to controls while the 49,XXXXY group had similar or somewhat reduced CT relative to controls.

Discussion: The current study provides the first vertex-level descriptions of CT and CSA in youth with DS and 49,XXXXY. Despite the fact that both disorders are characterized by reduced cortical volume, dissociations were observed with regard to CT- namely, the DS group had increased CT particularly in anterior and posterior brain regions and the 49,XXXXY group had similar or decreased CT in these regions. In addition to cortical volume reductions in DS, cerebellar volume was reduced. (These results are forthcoming for 49,XXXXY). Taken together, these findings provide evidence for areas of neuroanatomic overlap and divergence for DS and 49,XXXXY and suggest that there may be some shared targets for biomedical and educational interventions as well as syndrome-specific targets.

Key References:
Blumenthal et al. (2014). Neuroimage Clinical, 2: 197-203.
Symposia Title: Using Structural MRI to Identify Shared and Unique Neuroanatomic Vulnerabilities in Five Developmental Disorders

Chair: Nancy Raitano Lee

Paper Title: Longitudinal Cortical Development During Adolescence and Young Adulthood in Autism Spectrum Disorders: Increased Cortical Thinning but Comparable Surface Area Changes

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Introduction: Prior reports suggest that autism spectrum disorder (ASD) is associated with atypically excessive early brain growth. Recent cross-sectional studies indicate that later cortical development during adolescence/adulthood might also be aberrant, though longitudinal designs are required to evaluate atypical growth trajectories. In order to address this open question, the present study sought to examine longitudinal changes in the two components of cortical volume, cortical thickness and surface area, among adolescents and young adults with ASD.

Methods: Two (70 total) high-resolution anatomic magnetic resonance imaging scans approximately two years apart were acquired from 17 higher functioning adolescents/young adults with ASD and 18 typically developing (TD) adolescents/young adults matched on age (ASD scan 1 mean=17.37 + 2.41, scan 2 mean=19.12 + 2.51; TD scan 1=17.46 + 1.45, scan 2=19.60 + 1.61), IQ (ASD mean=116.59 + 13.05; TD mean=116.17 + 9.54), sex ratio (male:female - ASD=15:2; TD=17:1), and handedness (right:left - ASD=15:2; TD=16:2). The FreeSurfer image analysis (version 5.1) suite was utilized to derive vertex-level cortical thickness and surface area values and to complete longitudinal analyses.

Results: There was widespread accelerated cortical thinning for the ASD group as compared to the TD group that was most prominent in two areas of the left hemisphere, the posterior portion of ventral temporal cortex and superior parietal cortex (cluster corrected p<.01). This greater longitudinal cortical thinning was driven by significantly thinner cortex in these regions in ASD at time 2 (ps<.01). Furthermore, linking these structural abnormalities with behavior in ASD, cortical thinning in these regions was associated with increased parent ratings of everyday executive function impairments and greater ASD social symptom severity. In contrast to cortical thickness, surface area was not found to differ in its longitudinal growth patterns when comparing the ASD and TD groups during adolescence/young adulthood.

Discussion: The present longitudinal study complements and extends prior cross-sectional research by demonstrating extended cortical thinning in ASD during adolescence and into young adulthood. Specifically, in portions of the temporal and parietal lobes, thinning of the cortex appears to have slowed in TD individuals, while this process continues to occur in these regions in individuals with ASD during this developmental window. Furthermore, this cortical thinning is associated with everyday executive functioning impairments, which have been consistently noted in the extant literature, along with ASD social symptomatology. Examination of differences in growth patterns of cortical surface area, however, did not reveal a similar pattern of findings: TD and ASD individuals exhibited comparable longitudinal changes in surface area during this age range. Taken together, these results provide further evidence for a second period of atypical cortical development in ASD marked by increased cortical thinning in late adolescence/young adulthood.
Symposia Title: Using Structural MRI to Identify Shared and Unique Neuroanatomic Vulnerabilities in Five Developmental Disorders

Chair: Nancy Raitano Lee

Paper Title: Distinct Regions of the Cerebellum Show Grey Matter Decreases in Autism, ADHD, and Developmental Dyslexia

Author(s): Catherine J. Stoodley, American University

Introduction: Our understanding of the human cerebellum has changed significantly in the past 20 years. Traditionally considered a motor structure, anatomical, clinical, and neuroimaging data have converged to suggest that the cerebellum has a role in the modulation of cerebro-cerebellar circuits involved in cognition and emotion as well as motor control. This role beyond motor control is supported by anatomical connections between the cerebellum and prefrontal and parietal association areas of the cerebral cortex. Further support comes from data indicating that the cerebellum shows resting-state functional connectivity with fronto-parietal, dorsal and ventral attention, default mode, and limbic networks. Within the cerebellum, different regions are involved in overt motor control vs. cognitive and emotional processing, giving rise to a functional topography within the cerebellum. This cerebellar functional topography is based on the anatomical connections with the cerebral cortex and spinal cord: lobules I-V and lobule VIII are predominantly sensorimotor; lobules VI and VII form circuits with frontal and parietal association cortices; lobule IX may participate in multiple cortical networks, including the default mode network; and lobule X comprises the vestibulocerebellum. Cerebellar functional topography is important when considering the role of the cerebellum in developmental disorders. Neuroimaging studies have reported cerebellar structural and functional differences in autism spectrum disorder (ASD), attention deficit hyperactivity disorder (ADHD), and developmental dyslexia. However, it is not clear whether the same or different cerebellar regions are affected in these disorders. The convergence and divergence of cerebellar structural differences in ASD, ADHD and dyslexia can indicate whether specific to cerebro-cerebellar circuits are affected in each disorder or whether cerebellar differences represent a more general characteristic of these developmental disorders.

Methods: We conducted an anatomic likelihood estimate (ALE) meta-analysis on voxel-based morphometric (VBM) studies which compared ASD (17 studies), ADHD (10 studies), and dyslexic (10 studies) participants with age-matched typically-developing controls. A second ALE analysis included additional studies in which the cerebellum was a region of interest (ROI).

Results: The results revealed no cerebellar regions of significantly increased grey matter (GM) in ASD, ADHD or dyslexia. In ASD, clusters of reduced GM were found in the inferior cerebellar vermis (lobule IX), left lobule VIII B and right Crus I. In ADHD, significantly decreased GM was found bilaterally in lobule IX, whereas participants with developmental dyslexia showed GM decreases in left lobule VI. There was no overlap between the cerebellar clusters identified in each disorder. To evaluate the potential functional significance of the regions revealed in both whole-brain and cerebellar ROI ALE analyses, we used Buckner and colleagues’ 7-network functional connectivity map available in the SUIT cerebellar atlas. The cerebellar regions identified in ASD showed functional connectivity with frontoparietal, default mode, somatomotor, and limbic networks; in ADHD, the clusters were part of dorsal and ventral attention networks; and in dyslexia, the clusters involved ventral attention, frontoparietal, and default mode networks.

Discussion: These results suggest that different cerebellar regions are affected in ASD, ADHD, and dyslexia. Further, the clusters where anatomical differences were found in each disorder are associated with different functional circuits, and the affected cerebro-cerebellar circuits are consistent with the behavioral profiles of each disorder: e.g., the default mode network in ASD, the dorsal attention network in ADHD. These findings indicate that the specific sub-region of the cerebellum that is affected in a given developmental disorder should be considered in the context of cerebellar functional topography and cerebro-cerebellar connectivity. Future studies will aim to determine specific contribution of cerebellar dysfunction to the etiology and behavioral manifestations of autism, ADHD, and developmental dyslexia.
Participant ID: 17

Symposium Title: Biomarkers of Mild Cognitive Impairment and Alzheimer’s Disease in Adults With Down Syndrome: Insights Gained With the Use of ‘Big Data’

Chair(s): Wayne Silverman, Kennedy Krieger Institute, Johns Hopkins University School of Medicine

This symposium will present data from a large transdisciplinary longitudinal study of aging and dementia in adults with Down syndrome. We will discuss the neuropsychological and genetic biomarkers of mild cognitive impairment and Alzheimer’s disease.
**Symposia Title:** Biomarkers of Mild Cognitive Impairment and Alzheimer’s Disease in Adults With Down Syndrome: Insights Gained With the Use of ‘Big Data’

**Chair:** Wayne Silverman

**Paper Title:** Clinical Subtypes of Mild Cognitive Impairment in Adults With Down Syndrome

**Author(s):** Sharon J. Krinsky-McHale, New York State Institute for Basic Research in Developmental Disabilities  
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Wayne Silverman, Kennedy Krieger Institute, Johns Hopkins University School of Medicine

**Introduction:** Mild Cognitive Impairment (MCI) is defined clinically as decline intermediate between changes associated with brain aging, per se, and those that occur with dementia [1]. Early criteria for MCI were specific to isolated memory deficits but it has now become clear that the clinical manifestations of MCI are heterogeneous and several subtypes have been identified depending on the nature of impairment [1,2]. The aim of this study was to determine if there are specific patterns of cognitive profiles in adults with Down syndrome with MCI and whether this distinction has prognostic value for the progression to dementia.

**Participants:** All individuals were enrolled in a multidisciplinary longitudinal study focused on aging and dementia [3]. Their trisomy 21 status has been confirmed by karyotyping. For the present study, only adults with Down syndrome and MCI (N=126) were examined (Mage =55.6 at diagnosis, range 46 to 78 years; MIQ =34.2, range 20 to 53). Other inclusion criteria included completing at least two cognitive tests in our neuropsychological battery and participating in at least two cycles of assessment (see below) one of which had to be subsequent to their MCI diagnosis.

**Procedure:** All participants received comprehensive evaluations at approximately 18-month intervals. These included direct assessment of selected cognitive functions, informant interviews, and review of medical records [3]. Following each assessment cycle the dementia status of each participant was rated at a Consensus Conference based upon consideration of all information available [4]. Consistent with current definitions of MCI subtypes for the general population, we classified participants into four subtypes [2], based on differences in the number and type of impaired cognitive domains: amnestic MCI single domain (aMCI-s) if there was an objective impairment in memory but not in any other domain of cognitive functioning, amnestic multiple domain (aMCI-m) if at least one other cognitive domain was impaired, non-amnestic MCI single domain (naMCI-s) if a single non-memory domain was impaired and non-amnestic MCI multiple domain (naMCI-m) if at least two non-memory cognitive domains were impaired.

**Results:** Subtypes of MCI differed in frequency, $\chi^2 (3, n=126) =112.3, p<.001$. Amnestic multiple domain (aMCI-m) was clearly the most frequent subtype (n = 81, 64.3%). Frequencies for other subtypes were: aMCI-s (n=28, 22.2%), naMCI-s (n=12, 9.5%) and naMCI-m (n=5, 4.0%). Approximately 2½ years following their MCI diagnosis, 87 (69%) individuals had shown further declines consistent with dementia however, the tendency to convert to dementia did not differ by MCI subtype.

**Discussion:** Using a detailed neuropsychological battery, this study found that memory decline is the most prominent symptom of MCI in adults with Down syndrome and that it seems to constitute an early stage in the progression to dementia (cf:[5]). Classification of individuals with Down syndrome into subtypes does not appear to have prognostic value above and beyond the MCI diagnosis.

**Key References:**


**Acknowledgments:** This work was supported by funds from the New York State Office for People with Developmental Disabilities and NIH grant P01 HD35897 (Silverman).
**Symposia Title:** Biomarkers of Mild Cognitive Impairment and Alzheimer’s Disease in Adults With Down Syndrome: Insights Gained With the Use of ‘Big Data’

**Chair:** Wayne Silverman

**Paper Title:** Variants on Candidate Genes Contribute to Age-At-Onset of Alzheimer’s Disease in Adults With Down Syndrome

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**Introduction:** Alzheimer disease (AD) is the most common form of dementia; yet, there is no effective treatment. The neuropathological changes of AD begin decades before clinical symptoms at a time when disease treatments are most likely to be effective. Understanding factors associated with age at onset (AAO) of AD can identify pathways that increase risk for development of AD and help to develop interventions for high-risk individuals. To address this issue, this study examined adults with Down Syndrome (DS) who are at high risk of developing AD. Previous studies have provided evidence for genetic contribution to the risk of AD in the general population. However, the role of these genetic factors has not been well studied in adults with DS. Because of trisomy 21, adults with DS overexpress genes on chromosome 21 including the gene for β-amyloid precursor protein (APP), which is associated with early onset of AD neuropathology and high risk for dementia. However, there is significant individual variation in AAO, and not all adults with DS develop AD.

**Methods:** We studied 313 adults with DS who were ascertained through the developmental disability service and followed for ~4.3 years. Cognitive assessments, caregiver interviews, medical record reviews, and neurological examinations were used to classify AD. We examined SNPs on candidate genes that have been previously implicated in the DS or general populations. For this purpose, the Illumina GoldenGate custom array was used. To assess how genotype contributes to AAO of AD, we applied Cox proportional hazard model that assumed a dosage model and controlled for sex, level of intellectual disability and presence/absence of APOE ε4 allele. In Stage 1, we pruned SNPs to generate tag SNPs so that the multiple-testing penalty is minimized, while these genes are covered sufficiently. In Stage 2, genes that had ≥1 SNP reaching a threshold p-value<0.05 were further studied using all SNPs. Gene-wise adjusted p-value was computed using the Benjamini and Hochberg method. Analysis was performed in PLINK or R.

**Results:** Stage 1 scan identified SNPs in DSCR1, RUNX1, BACE2, and APP on chromosome 21 and SNPs in MSRA, CST3, PCHD11X, IDE, CLU, ZNF224, PICALM, SORCS1, ALDH18A1, NEDD9, and DAPK1 on chromosomes other than 21. In Stage 2, all SNPs on genes that screened positive in stage 1 were analyzed using the same model. Using gene-wise adjusted p-value<0.05, multiple SNPs in DSCR1 (HR=1.59-1.9), CST3 (HR=1.69-2.16), and IDE (HR=0.528) were significantly associated with AAO of AD. Broadly classified, SNPs in these genes may influence central nervous system development (DSCR1); APP processing since Aβ and insulin compete with IDE (IDE); and cardiovascular pathway toward AD (CST3).

**Conclusions:** We identified SNPs in DSCR1, CST3 and IDE that were associated with AAO. The findings from this study extend our earlier studies which showed that these genes were associated with variation in levels of Aβ (IDE) and increased risk of AD (CST3). We are now examining the exome to identify coding SNPs that may have contributed to alteration in AAO in these genes and elsewhere in the exome.

**Acknowledgments:** Supported by grants R01AG014673 (Schupf) and P01HD35897 (Silverman) from NIA and NICHD and by NYS through its Office for People with Developmental Disabilities.
Introduction: Adults with Down Syndrome (DS) carry three copies of the gene for β amyloid precursor protein (APP), and they have an increased risk of dementia; yet, age at onset of AD varies widely among individuals with DS. Previous genetic studies in the general population have identified over 20 genes that contribute to the risk for Alzheimer disease (AD). We and others have examined the role of genetic factors in adults with DS, and have reported several genes that were associated with DS-AD and related traits. Those genes included candidate genes on chromosome 21 as well as genes on chromosomes other than 21. In the present study, we extended our earlier fine mapping efforts by performing exome sequencing to identify variants that may have functional relevance in risk of dementia.

Methods: We previously reported that SNPs on the CST3 and MARK4 genes are associated with AD in adults with DS, where the CST3 gene may influence formation of amyloid fibril and the MARK4 gene may be involved in microtubule regulation. To examine these genes in greater detail and also explore variants elsewhere in the exome, we sequenced the exome in 46 adults with DS from a total of 357 who had been previously genotyped for the fine mapping study. Study participants were ascertained through the developmental disability service systems of New York and neighboring states and were followed at 14-18 month intervals for ~5 years. Cognitive assessments, caregiver interviews, medical record reviews and neurological examinations were used to classify dementia. Twenty-three individuals had AD, and 23 did not. None of the sequenced individuals had a copy of APOE ε4 allele. Exome sequencing was performed using the Illumina HiSeq platform. In the present exome sequencing experiment, we focus on genes that are on chromosomes other than 21. Following quality control filtering, allelic association was performed using PLINK and R.

Results: Exome sequencing yielded 229,168 variants (215,890 SNPs and 13,278 indels). After excluding homozygotes and low quality variants, we analyzed 206,197 autosomal SNPs from non-chromosome 21. For the previously identified CST3 gene on 20p11.21 and the MARK4 gene on 19q13.32, a SNP with the strongest effect size was observed in MARK4 (OR=5.18); however, the strength of association did not reach statistical significance. For the other regions of the exome, a strong signal was observed in the DACT1 gene on 14q23.1 (p=0.0015), which may be involved in organization of synapse, formation of dendrite formation, and establishment of spines.

Conclusions: The present study identified coding SNPs that may contribute to elevated risk of dementia is adults with DS. These findings further demonstrate that multiple genes on chromosomes other than 21 are likely to influence variation in risk for AD. Because these genetic variants are rare, the present study with small sample size serves as a discovery experiment that will need to be validated in a larger set of adults with DS as well as in the general population.

Acknowledgments: Supported by grants R01AG014673 (Schupf) and P01HD35897 (Silverman) from NIA and NICHD and by NYS through its Office for People with Developmental Disabilities.
**Symposium Title:** Innovative Frameworks for the Study of Children With Autism Spectrum Disorder

**Chair and Discussant:** Jason K. Baker, California State University-Fullerton

This symposium includes four presentations from early career researchers who are applying innovative frameworks to the study of children with autism spectrum disorder (ASD). Each of the areas discussed has demonstrated importance to child development, and several of the presentations represent collaborations between ASD researchers and experts in the study of these topics in neurotypical populations. Two projects focus on the psychophysiological reactivity of children with ASD. Schwartzmann et al. consider status-group differences in the experience of certain emotions by children with ASD (via respiratory sinus arrhythmia) in order to better tailor emotion-based interventions to this population. Fenning et al. also examine psychophysiological arousal (electrodermal activity), but with an eye towards improving our understanding of individual differences in the emotional and behavioral functioning of children with ASD. This study also integrates psychophysiological measurement with traditional emotion regulation frameworks, popular in the study of child development, to understand heterogeneity in ASD. Obesity and transactions between physical health and child adjustment are important developmental concerns that are arguably even more critical to consider for children with ASD and intellectual/developmental disabilities. Davis, Sanner, and Neece report on a pilot study in which they adapt an established treatment program for obesity to these populations. Finally, Hartley and colleagues apply a family systems perspective to ASD, not only to understand potential cascading effects of children's functioning on the marital relationship, but also to consider bidirectional effects from these less proximal family relationships back to the children with ASD. Dr. Baker will provide a brief discussion of the processes by which frameworks developed with the general population are often applied to more specific groups, proposing that we are still in the very early stages of investigating these areas in ASD, and underscoring the importance of following these lines of research as they continue to develop.
Symposia Title: Innovative Frameworks for the Study of Children With Autism Spectrum Disorder

Chair: Jason K. Baker

Paper Title: Psychophysiology of Emotional Reactivity in Children With ASD

Author(s): Ben Schwartzman, University of California Los Angeles
Jeffrey Wood, University of California Los Angeles
Sheila Crowell, University of Utah
Yue Yu, Indiana University, Purdue University Indianapolis

Introduction: Respiratory Sinus Arrhythmia (RSA) is a naturally occurring variation in heart rate that occurs during the breathing cycle controlled by the parasympathetic nervous system (Porges, 1995). As described in Porges' Polyvagal Theory, RSA has been theorized as a biomarker of emotion regulation which, in turn, affects overall social functioning (Porges, 1995). Given the social difficulties associated with Autism Spectrum Disorder (ASD), measuring RSA in this population should provide valuable insights into the biological mechanisms underlying these deficits. Previous studies have shown that higher RSA was an indicator of better social functioning in children and adolescents with ASD (Neuhaus et al., 2013, Patriquin et al., 2013, Bal et al., 2010, and Van Hecke et al., 2009).

Methods: A sample of 80 children (40 ASD, 40 typical-developing), aged 6 to 13, participated the study. Eligibility for children with ASD was established with Autism Diagnostic Interview-Revised (Lord et al.1994) and Autism Diagnostic Observation Schedule (Lord et al, 2003). RSA data were collected while participants watched baseline videos (videos of fish swimming accompanied by relaxing music) and videos of characters experiencing different emotions (Angry, Scared, Sad, Embarrassed, and Happy). In the paradigm, psychophysiological signals are collected using the Biopac M150 base unit. To measure respiratory sinus arrhythmia (RSA), one Ag-AgCl electrode is placed on the left, lowermost rib, and a second Ag-AgCl electrode is placed on the middle of the right collarbone. After the psychophysiological data is acquired and recorded using Biopac AcqKnowledge version 4.2, it is analyzed using MindWare HRV 3.0.22. HRV 3.0.22 is used to calculate the RSA data in 30-s epochs, and an average RSA score is determined for each of the different emotion trials.

Results: A significant difference emerged between ASD and TD participants from baseline to emotion video change scores for the Embarrassed (ASD=-.095 and TD=-.733 at p=.008) and Sad (ASD=-.147 and TD=-.634 at p=.045) videos. Results were not significant for the other three emotion videos when comparing groups.

Discussion: This result is somewhat consistent with previous findings that RSA functions differently in children with ASD as compared to typically developing children. Though we did not find differences in baseline RSA level between the groups as found in previous research, we found significant differences between groups in average RSA change from baseline to emotion videos. Children with ASD, as compared to their typically developing peers, may physiologically experience sadness and embarrassment differently than their typically developing peers. These results highlight the potential of exploring biological measures of social functioning when evaluating interventions for children and adolescents with ASD.

Key References:


**Symposia Title:** Innovative Frameworks for the Study of Children With Autism Spectrum Disorder  
**Chair:** Jason K. Baker  
**Paper Title:** Electrodermal Activity, Autism Symptoms, and Emotion Regulation in Children With ASD  
**Author(s):** Rachel M. Fenning, California State University-Fullerton  
Jason K. Baker, California State University-Fullerton  
Stephen Erath, Auburn University  
Mariann Howland, California State University-Fullerton  
Jacquelyn Moffitt, California State University-Fullerton

**Introduction:** Understanding heterogeneity in autism spectrum disorder (ASD) is of great interest to the field. It is likely that factors identified as important to the development of problems and competence in neurotypical populations can inform efforts to understand individual differences in the emotional and behavioral functioning of children with ASD. Children’s ability to regulate their emotions is considered a foundational skill, but emotion regulation frameworks have not been applied to the study of ASD until recently. Similarly, psychophysiological arousal tendencies are related to broad indices of functioning in non-ASD populations. Research on electrodermal activity (EDA), an index of sympathetic nervous system arousal, has been studied in ASD in order to investigate potential status-group differences, but not yet to explain heterogeneity among children with ASD. The current pilot study examined associations between EDA, autism symptoms, and emotion regulation in children with ASD.

**Methods:** A culturally diverse sample of 25 children (20 males) ages 4 to 11 (M= 6.56, SD= 1.70) with an ASD diagnosis participated in a laboratory visit with their primary caregivers. The two-hour visit included administration of the Autism Diagnostic Observation Schedule-2 (ADOS-2), the Stanford-Binet 5 Abbreviated IQ Battery, and several structured tasks including a locked-box, child-alone frustration task, and a parent-child problem-solving task. The ADOS-2 comparison score was used to measure ASD symptoms. Emotion dysregulation was coded from video of the two structured tasks using the reliable and valid Emotion Dysregulation Coding System (Baker et al., 2007). Electrodermal activity was obtained through wireless sensors worn by the children throughout the visit, which recorded EDA, temperature, and movement at eight hertz.

**Results:** No demographic variable considered was related to the variables of interest in a manner that would confound the findings. Dysregulation scores for the child-alone and parent-child tasks were correlated, $r = .43$, $p < .05$, and were combined to form a single dysregulation composite. Due to the traditionally non-normal distribution of EDA data, non-parametric analyses and transformation into ranked scores were used for these data. The mean level and average variability (e.g., SD) of EDA scores were highly correlated, $rs = .86$, $p < .001$, thus variability was prioritized. Higher rates of emotion dysregulation were related to higher EDA variability, $rs = .54$, $p < .01$, more autism symptoms, $r = .49$, $p < .05$, and lower IQ, $r = -.53$, $p < .01$. IQ was not significantly related to EDA, $rs = -.18ns$, or autism symptoms, $r = -.34ns$, and regression suggested that it was independently associated with dysregulation, beta= -.38, $p < .05$. EDA and autism symptoms were highly related, $rs = .62$, $p < .01$, and regression suggested that it was the shared variance of these factors that was important to dysregulation, $R^2 = .32$, $p < .05$.

**Discussion:** These pilot findings suggest that children’s cognitive level, physiological arousal tendencies, and autism symptoms are important for understanding emotion regulation abilities in ASD. Studies involving larger samples are necessary to replicate these findings, to further investigate potential mediation involving EDA and autism symptoms in the prediction of dysregulation, and to consider interactions between children’s biological systems and environmental influences.

**Key References:**


**Introduction:** Children and adolescents with autism spectrum disorders (ASD) and intellectual and developmental disabilities (IDD) show elevated rates of overweight and obesity compared to their typically developing peers. The increased rates of overweight and obesity in these populations have important implications, as higher body mass index (BMI) is related to numerous negative health outcomes. Despite the negative consequences of obesity, interventions targeting weight loss and health behaviors in children and adolescents with IDD and/or ASD have been limited. In the current study, we examined the characteristics of children diagnosed with ASD compared to those with other forms of IDD in a sample of children whose weights were categorized as overweight or obese. Additionally, we assessed the feasibility of conducting a day camp intervention targeting physical fitness and health behaviors for this sample.

**Method:** Operation FIT is a day camp intervention that was established to improve the physical health of typically developing children and adolescents who are overweight or obese. The overall aims of the camp are to increase nutrition-related knowledge and physical activity levels. Throughout each day of camp, educational and physical activities are alternated. Additionally, children practice skills to increase healthy behaviors, such as reading nutrition labels. The camp was held over four consecutive nine-hour days and included 29 children, ages 9 to 15 years old, with ASD and/or IDD. This paper will report findings from the first Operation FIT program adapted for children with IDD.

**Results:** Eighty-one children were screened, 45 were eligible for the camp, 29 children enrolled in the camp. The majority of participants were male (66.7%) and the sample was diverse in terms of ethnicity (55.6% Hispanic, 22.2% African American, 18.5% Caucasian, and 3.7% Asian) and income (71.4% in poverty). The mean age was 12.04 years (SD=1.89), and 55.2% of participants were diagnosed with ASD, 44.8% were diagnosed with other forms of IDD. Most of the children were categorized as obese (72%), and there was no significant difference in weight category between children with ASD and those with other IDDs (p>.05). On average, children with ASD participated in fewer physical activities than children with other IDDs (M=1.73, M=1.84, respectively) and engaged in these activities for fewer hours per week (M=3.22, M=3.90, respectively), though these differences were not significant. In the overall sample, 50.0% of children exhibited clinical levels of behavior problems and there was no significant difference between children with ASD and other forms of IDD. Levels of behavior problems predicted increased BMI, such that children who exhibited clinical levels of behavior problems showed a higher BMI compared to children with borderline or non-clinical levels of problem behaviors (r=.40, p<.05). Regarding the feasibility of the intervention, we had a low attrition rate (3.4%). The average number of days attended was 3.52 (SD=1.09) and 79.3% of the participants attended every session. Additional participant characteristic comparisons and feasibility data will be presented and preliminary intervention outcomes will be discussed.

**Discussion:** The participants in the current study showed high levels of risk for both proximal and distal negative health and psychosocial outcomes. There was no significant difference between children with ASD and those with IDD when risk factors were compared. In this sample, clinical levels of behavior problems were shown to be a risk factor for increased BMI, which is an important finding given the elevated behavior problems consistently reported in this population. Based on the preliminary recruitment, attrition, and attendance data, a camp-based format appears feasible as an intervention strategy for behaviors and knowledge related to overweight and obesity in this population. Limitations to the current study and directions for future research will also be discussed.
Introduction: Children with autism spectrum disorders (ASD) present with a challenging profile of autism symptoms and often display co-occurring emotional and behavioral problems (e.g., inattention and anxious affect), which can be stressful for parents. There is theoretical and empirical evidence to suggest that chronic parenting stress affects marital adjustment. In turn, parents' marital adjustment has been shown to lead to increases in child emotional and behavioral problems in research on the general population. We will discuss data from an ongoing longitudinal study examining the within-family day-to-day associations between parents' marital interactions and the emotional and behavioral problems of children with ASD.

Methods: Analyses included 175 families of children (aged 5-12 yrs) with ASD. All children had a documented diagnosis of ASD by an educational or medical specialist, which included the Autism Diagnostic and Observational Schedule. Parents had a mean age of 37.45 yrs (SD = 3.52) and median household income of $70 to $79K. Children with ASD had an average age of 8.81 yrs (SD = 1.53) and most were male (76.6%). Mothers and fathers independently completed a 14-day daily diary in which they reported on 7 positive marital interactions (e.g. sharing a joke), 7 negative marital interactions (e.g., critical comment), and the occurrence of marital disagreements on each day. Parents also independently completed the Scales of Independent Behavior-Revised each day to assess the frequency x severity of child emotional and behavioral problems.

Results: Multilevel multivariate modeling using HLM was conducted to examine within-person same-day associations between child emotional and behavioral problems and marital interaction variables (Positive Marital Interaction, Negative Marital Interaction, and Frequency x Severity of Marital Disagreements) within the 14-day daily diary. The between-couple effects of family, parent, and child variables on initial status of marital interaction variables and their interaction with within-person predictors were examined. Child emotional and behavioral problems significantly co-varied with the Negative Marital Interaction score and the Frequency x Severity of Marital Disagreement score. The mean level of child emotional and behavioral problems (between-couple) significantly interacted with daily child emotional and behavior problems (within-person) in models. In order to begin to elucidate causal pathways, analyses were also conducted to examine within-person associations between previous-day child emotional and behavioral problems and next-day marital interaction variables. Vice versa, the association between previous-day marital interaction variables and next-day child emotional and behavioral problems were examined.

Discussion: Findings offer insight into the unfolding of 'real time' day-to-day associations between marital interactions and the child with ASD’s emotional and behavioral problems in their natural and spontaneous context. Results suggest that within-parents, negative couple interactions and marital disagreements fluctuate together with changes in child emotional and behavior problems. Findings also suggest that parents of children with ASD who typically have a lower level of behavior problems are more influenced by a ‘bad parenting’ day (i.e., lots of child behavior problems) than are parents of children with ASD who typically have a higher level of emotional behavior problems. Child, parent, and family factors related to marital interactions will also be discussed. Evidence suggests that there is a bi-directional causal pathway between the day-to-day marital interactions of parents and the emotional and behavioral problems of children with ASD. Implications for interventions and next steps in research will be discussed.

Key References:

Symposium Title: Clinical Research Issues in the Study of Pain and Intellectual and Developmental Disabilities

Chair: Frank Symons, University of Minnesota
Discussant: Ken Craig, University of British Columbia

Pain is a universal phenomenon causing tremendous human suffering and compromising the quality of life for countless individuals. The U.S. congress had declared the prior decade 2001-2010 as the Decade of Pain Control and Research, however we estimate a decade devoted to pain research yielded fewer than 80 empirical papers about pain in individuals with intellectual and developmental disabilities (I/DD). There has been little change in the past five years. And yet, there is no good reason to believe that pain is any less frequent in an individual living with an I/DD or that such an individual would be insensitive or indifferent to pain. Numerous functional limitations as well as the underlying neurologic condition itself frequently confound the presentation of pain. Regardless of the degree of the disability, however, pain is often a part of daily life for individuals with I/DD. In this symposium, we have assembled four unique datasets spanning beliefs to behavior to biology relevant to pain signs and symptoms in samples of individuals with I/DD. The talks will focus on approach and assumptions as much as findings and outcomes with discussions oriented around remaining issues and the next steps toward building sustained programs of research specific to pain in I/DD.
Purpose: Individuals with intellectual disabilities (ID) often receive care from a variety of different caregivers. In these circumstances, primary caregivers are often unavailable to assist with pain assessment and management decisions. No research has examined the beliefs of respite workers. The purpose of this study was to (1) compare disability and pain-related beliefs in respite workers (RW) versus undergraduate students (UG) with limited experience with children with ID and (2) determine whether beliefs can affect pain assessment and management decisions.

Methods: Participants consisted of 217 UG (Mage = 19.63) and 56 RW (Mage = 33.37). Participants read and responded to six situational vignettes [four adapted from previous research] designed to measure care decisions. Participants also completed: the Pain Opinion Questionnaire, measuring pain beliefs regarding children with ID versus ‘typically developing’ children, as well as the Mental Retardation Attitudes Inventory - Revised measuring individual attitudes towards those with disabilities.

Select Results: RW held more positive disability-related beliefs than the UG sample, t(267) = 4.72, p < .001. RW were also more likely to report that children with severe ID sense less pain than children without ID compared to UG, t(262) = 2.63, p = .009. Compared to UG, RW believed that a higher percentage of children with both moderate, t(263) = 2.08, p = .04, and severe ID, t(261) = 2.51, p = .01, demonstrate a lower emotional reaction to pain than ‘typically developing’ children. Participant beliefs and level of involvement with children with ID who were nonverbal were significantly related to care decisions across vignettes (all p’s < .05). Multiple regressions indicated that participants’ disability and pain-related beliefs significantly predicted care decisions. Specifically, disability-related beliefs significantly predicted ratings for need for medical attention, β = -.197, t(242) = -3.76, p < .001 and need for other attention, β = .140, t(251) = 2.37, p = .018. Further, participant’s ratings for children with mild ID and their ability to sense pain compared to children without ID also predicted ratings for need for medical attention, β = .200, t(242) = 2.75, p = .006.

Discussion: Respite workers hold different beliefs than those of UG with little to no experience with children with ID. Some beliefs held by RW may not accurately reflect pain experience in children with ID. Interaction and experience with children with ID may contribute to these beliefs which in turn could impact pain assessment and management in respite care settings, and ultimately, children’s health-related quality of life.

Key References:


Symposia Title: Clinical Research Issues in the Study of Pain and Intellectual and Developmental Disabilities

Chair: Frank Symons

Paper Title: Assessment of Sensitivity to Pain in Prader-Willi Syndrome

Author(s): Griffin Rooker, Johns Hopkins School of Medicine, Kennedy Krieger Institute
Brian Iwata, University of Florida
Erin Camp, Little Leaves Behavioral Services

Purpose: Self-injurious behavior (SIB) is a common behavioral feature of Prader-Willi syndrome (PWS). But, the relation between pain and SIB in PWS has not been investigated. More generally, there has been only one study on the nature of pain and pain responding in PWS1. The purpose of this study was to begin investigating pain function in PWS and the relation of the pain response to SIB.

Methods: We examined pain-sensitivity of individuals diagnosed with Prader-Willi Syndrome (PWS) by applying the cold pressor test to three samples: typically developing individuals, individuals diagnosed with PWS, and individuals who diagnosed with PWS who engaged in SIB. In addition, each subject was tested twice to examine the stability of the findings.

Results: Results indicated that individuals diagnosed with PWS were more sensitive to painful stimulation than typically developing individuals, and that individuals diagnosed with PWS who engaged in self-injury were the most sensitive to painful stimulation. Furthermore, individuals diagnosed with PWS decreased the latency to removal in most cases during the second test; whereas the opposite was true for typically developing individuals. Results from behavioral assessments indicated that SIB was maintained by non-social consequences in all cases.

Discussion: As far as we know, this was the first study to characterize pain function in PWS using the cold-pressor test. Results this study suggest that on-going pain responding is intact in individuals with PWS and for the subgroup with SIB, and that these individuals had lower thresholds. The potential relation between non-social SIB and pain is discussed.

Key References:

Symposia Title: Clinical Research Issues in the Study of Pain and Intellectual and Developmental Disabilities

Chair: Frank Symons

Paper Title: Comparing Cutaneous Sensory-Tactile Reactivity Between Children With and Without Global Developmental Delay

Author(s): Chantel Barney, Gillette Children's Specialty Healthcare
Raymond Tervo, Mayo Clinic
Frank Symons, University of Minnesota

Purpose: The aim of the investigation was to compare gender and age-matched children with and without global developmental delay on behavioral reactivity to an array of calibrated tactile sensory stimuli. Assessing pain in children with developmental delays remains a challenge, particularly in the presence of any associated intellectual or communicative deficits. To date, there has been very little work investigating early pain or sensory reactivity profiles of children with developmental delay and no work directly comparing behavioral reactivity profiles in relation to 'healthy controls'.

Methods: A case control design was used to test whether 20 children with GDD (60% male; M age = 4.91 years, SD=1.13) would be more or less reactive to a sensory test compared to a control group of 20 typically developing children (60% male; M age = 3.49 years, SD=1.08). Sensory reactivity was indexed by vocal, facial, and body behaviors exhibited during the sensory test (behaviors were selected from existing non-verbal pain checklists and further operationally defined for direct observation coding by coders blind to child and stimulus status). Observers were trained to reliability across codes (> 80%).

Results: Children with GDD exhibited a significantly greater proportion of vocal (M=.33, SD=.30) and body (M=.48, SD=.25) behavioral reactivity compared to the proportion of vocal (M=.06, SD=.16; t(28.9)=3.47, p<.01) and body (M=.28, SD=.24; t(37)=2.46, p<.05) pain behaviors of children in the typical development control group. There were no significant differences in the duration of pain behaviors coded during the sham and heat stimulus trials. However, children with GDD exhibited, on average, significantly more pain behaviors during light touch (p<.05), pin prick (p<.01), cool (p<.05), pressure (p<.05), and repeated von Frey (p<.01) stimulus trials compared to the control group. For children with GDD, severity of self-injurious behavior significantly correlated with vocal (r=.58, p=.01) and body (r=.56, p<.05) pain behaviors exhibited during the pin prick trial.

Discussion: This study was the first to specifically measure the behavioral response of children with GDD to a calibrated sensory tactile test protocol and compare directly to a typically developing control group. The results of this study provide valuable albeit indirect information about the integrity of the sensory and nociceptive pathways of very young children with GDD and their ability to express pain behaviors in relation to an array of sensory tactile experiences. Given the directly observed reactivity, it would be fair to conclude that the tactile experience was being encoded centrally. The results provide further empirical evidence against the biased belief that individuals with developmental disability are insensitive or indifferent to pain. The result also suggest there are important unanswered questions regarding the apparent tactile hyper (not hypo) reactivity in the GDD sample.

Acknowledgements: This study was supported, in part, by Gillette Children's Specialty Healthcare Research Administration, and NIH Grant No. 44763 & 47201.
**Symposia Title:** Clinical Research Issues in the Study of Pain and Intellectual and Developmental Disabilities

**Chair:** Frank Symons

**Paper Title:** Characterizing Inflammatory Mediators in the Saliva and Cerebral Spinal Fluid of Children and Adolescents With Cerebral Palsy

**Author(s):** Frank Symons, University of Minnesota  
Chantel Barney, Gillette Children's Specialty Healthcare  
Samuel Roiko, Gillette Children's Specialty Healthcare  
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Michael Ehrhardt, University of Minnesota  
Angela Panoskaltsis-Mortari, University of Minnesota

**Purpose:** Cerebral palsy (CP) is a major neurodevelopmental disability associated with intrauterine infection and abnormal patterns of cytokines and growth factors that potentially predispose the development of CP. In children and adolescents with CP we aimed to examine 1) the inflammatory mediators in saliva and cerebral spinal fluid (CSF) specific to CP type and intensity, and 2) the association between inflammatory mediators and pain experience.

**Methods:** We undertook a clinically-oriented feasibility study to measure salivary and cerebral spinal fluid (CSF) levels of neuropeptides, cytokines, chemokines, and hormones relevant to nociceptive status from 34 pediatric patients with cerebral palsy (CP). Unstimulated (passively collected) saliva was collected using oral swabs and CSF was collected during intrathecal baclofen (ITB) pump implant or selective dorsal rhizotomy (SDR) surgical procedures. Parents reported pain type, frequency, duration, intensity, and pain interference with function for the 7-days prior to study enrollment.

**Results:** Participants (79.4% male; M age= 8.95 years, SD=4.65, range=4-18 years) had quadriplegia (n=23), diplegia, (n=9), and triplegia (n=2) types of cerebral palsy and were both ambulatory (Gross Motor Classification System [GMFCS] levels I-III; n=16) and non-ambulatory (GMFCS levels IV-V; n=18). Most children and adolescents with CP had experienced at least one episode of pain in the previous 7 days (91%) with musculoskeletal type pain being most frequent and gastrointestinal pain being most intense and long lasting. Thirty-eight analytes were measured in CSF and 35 in saliva. Analytes were characterized in terms of mean levels by pain severity, CP severity, and CP type. Salivary and CSF analytes were analyzed in relation to reported pain intensity and duration. CSF levels of Interleukin (IL)-6 and tumor necrosis factor (TNFa) differ significantly based on pain/no pain groups. CSF levels of IL-8 correlate with TNFa when pain is present ($p<.02$, $r=.93$) but not when pain is absent (ns, $r=.26$).

**Discussion:** This study is the first to characterize salivary and CSF neuropeptides, cytokines, chemokines, and hormones relevant to nociceptive status in children and adolescents with CP. The finding that some analyte levels were related to parent-reported pain experience demonstrates the possible clinical value of measuring salivary and CSF biomarkers relevant to nociception and inflammation. Further study is warranted to improve our understanding of molecular mechanisms related to the presentation of CP in an attempt to make a difference in the lives of children with developmental disabilities.

**Acknowledgements:** This study was supported, in part, by Gillette Children’s Specialty Healthcare Research Administration, the University of Minnesota’s Futures Program and NIH Grant No. 44763 & 69985.
Symposium Title: The Cutting Edge of Sequential Analysis Research: Simulations, Applications, and 'Big' Sequential Data

Chair: Blair Lloyd, Vanderbilt Kennedy Center, Vanderbilt University
Discussant: Jim Bodfish, Vanderbilt Kennedy Center, Vanderbilt University

In this symposium, we present three data sets aimed to inform sequential analysis research related to individuals with intellectual and developmental disabilities (I/DD). In the first presentation, we share results of a simulation study designed to compare methods of sequential analysis in terms of the accuracy of indices of association they produce. In the second presentation, we present an application of generalizability theory to sequential data on behavior-environment contingencies in classrooms to inform the number of observations necessary to produce reliable estimates. In the third paper, we present an application of time lag sequential analysis to high-resolution data on cardiac responsivity and behavioral reactivity for individuals with MeCP2 genetic syndromes. All three papers represent cutting edge approaches to sequential analysis in the field of I/DD.
Introduction: Sequential analysis is an approach combining measurement and statistical knowledge and theory to determine whether the occurrence of one event is associated with the subsequent occurrence of another event in one or more observations (Yoder & Symons, 2010). In the field of I/DD, sequential analysis methods have been applied to explore close temporal associations between variables ranging from parent-child social interactions, to potential reinforcing consequences following severe problem behavior, to physiological reactivity following stimulus presentations. A variety of methods exist to estimate sequential associations from direct observation data. Methods vary according to the behavior sampling approach, data representation, and indices of association calculated from 2x2 contingency tables. Three of the most common sequential analysis approaches include the event lag method, the time window method, and the concurrent interval method. One potential limitation of the concurrent interval method, however, is that the sequence of target events within interval is not preserved. Using simulated data, we conducted a comparison of four sequential analysis approaches: event lag, time window, concurrent interval, and a new interval method in which the target event sequence is preserved within interval.

Research Questions:
1. Across the four sequential analysis approaches, which approach produces the most accurate index of association?
2. Of the remaining approaches, which sequential analysis methods produce indices that are non-trivially different from the most accurate method?

Method: We generated 5,000 timed-event data streams, each lasting 3600 s (representing 1 hour of observation time). We randomly selected simple probabilities of each target event from uniform distributions to allow simple probabilities of each event to vary. We sequenced the events randomly to create a distribution of contingencies between Event 1 and Event 2 with a known mean contingency of zero. To estimate the contingencies, we applied the event lag and time window approaches to the timed event data streams, then imposed 10-s intervals over the timed-event streams to apply each concurrent interval approach. From the 2 x 2 contingency tables produced by each method of behavior sampling and data representation, we calculated operant contingency values (OCV) as the index of association. Indices closest to zero were considered most accurate given the randomly generated event streams. To address the second research question, we calculated effect sizes (Cohen's d) of the difference in estimated associations between the most accurate approach and each of the remaining approaches.

Results: Results indicated that the event lag method produced the most accurate indices of association (i.e., closest to zero). In addition, the modified concurrent interval method (i.e., preserving target event sequence within interval) was shown to produce sequential estimates that were non-trivially different from those produced by the event lag method, and revealed a negative bias.

Discussion: Results of the simulation study suggest using a timed-event behavior sampling approach with event pairs as coded units produces the most accurate estimates of sequential association. However, a challenge of using the event lag sequential analysis method in applied research is the identification of an exhaustive coding system in which all "relevant" events are included (Yoder & Symons, 2010). Though the event lag method may be most accurate under conditions of known exhaustive coding systems, other methods (e.g., concurrent interval) shown to produce sequential indices trivially different from the event lag method still may be most appropriate when the exhaustive coding system is unknown.

Key References:

Introduction: Generalizability theory (GT; Cronbach, Gleser, Nanda, & Rajaratnam, 1972) may be used to assess sources of variability within a set of measurement samples. Among other questions, GT may be applied to existing observational data sets to address how many observation sessions are necessary to yield reliable and representative estimates. Recently, the fields of education and psychology have seen an increase in G studies applied to a range of dependent measures, including curriculum-based measurement probes, behavior ratings, and systematic direct observation of on- and off-task behavior and social skills. GT has not, however, been applied to evaluate the reliability of behavior-environment contingencies or sequential associations. The purpose of this study was to evaluate the number of observations necessary to produce reliable estimates of antecedent-behavior contingencies and behavior-consequence contingencies for students with I/DD and severe problem behavior during 1:1 instruction in classroom settings.

Research Questions:
1. Are five 30-min observations sufficient to produce reliable estimates of contingencies between antecedent stimuli and problem behavior? If not, approximately how many observations would be necessary?
2. Are five 30-min observations sufficient to produce reliable estimates of contingencies between problem behavior and consequent stimuli? If not, approximately how many observations would be necessary?

Method: We conducted a minimum of five 30-min observations for seven students with I/DD and their paraprofessionals during the same 1:1 instructional period each day. We used a partial interval sampling procedure to code the occurrence of student problem behaviors and paraprofessional behaviors within an interval, preserving the temporal sequence of events in relation to problem behavior. For each behavior-environment contingency, we calculated Yule’s Q and operant contingency value (OCV; see Lloyd, Kennedy, & Yoder, 2013). We derived variance components using an ANOVA with Type III Sum of Squares to estimate person x session g coefficients (criterion g = .70). Finally, we used the variance component estimates to conduct decision (D) studies to determine the number of additional observations necessary to produce reliable contingency estimates.

Results: Results indicated that although five 30-min sessions were sufficient to produce reliable estimates of student problem behavior (g = .70), g coefficients for antecedent-behavior and behavior-consequence contingencies were variable, ranging from .17-.78. D studies indicated the number of 30-min observations required to produce reliable contingency estimates ranged from as few as 3 to as many as 10.

Discussion: Results suggest the amount of observational data necessary to yield reliable estimates of contingencies between two behaviors or events is likely to exceed the amount necessary to yield reliable estimates of each behavior in isolation. Results of this study should be interpreted with caution, however, given the small number of participants and given other methods of sequential analysis that exist and that may show different results. Future work in this area, including G studies applied to larger N samples and contingency estimates derived from different sequential analysis methods, are discussed.

Key References:
**Symposia Title:** The Cutting Edge of Sequential Analysis Research: Simulations, Applications, and 'Big' Sequential Data

**Chair:** Blair Lloyd

**Paper Title:** Sequential Analysis Integrating Cardiac and Behavioral Data During Sensory Testing in MeCP2 Genetic Syndromes

**Author(s):** John Hoch, University of Minnesota  
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**Introduction:** Rett Syndrome (RTT) results from under-expression of the gene MeCP2 and the corresponding reductions in the MeCP2 protein. Recently, MeCP2 Duplication (DUP), a syndrome resulting from over-expression of MeCP2 has been identified. There is little comparative work investigating the behavioral phenotypes of MeCP2 over- and under-expression. There are myriad documented autonomic regulatory issues in RTT but less is known about DUP. In this preliminary study, behavioral and cardiac reactivity to stimuli was examined during a quantitative sensory testing protocol using time lag sequential analysis and results were compared between groups that differed in MeCP2 gene impairment.

**Method:** Participants with Rett Syndrome (RTT; n=11), participants with MeCP2 Duplication (DUP; n=9), and typically developing preschoolers (TYP; n=16) were assessed using a sensory testing protocol involving brief application of stimuli (e.g. pin prick, pressure, heat). Participants' cardiac activity recorded during the assessments was synchronized with events coded from video of the sessions including participant vocalizations, facial or body movements and experimenter application of stimuli. Cardiac events were defined per individual as changes in heart rate above or below one standard deviation. These combined cardiac/behavioral event streams were analyzed for sequential dependencies between the sensory stimuli and cardiac or behavioral events occurring within five seconds of the stimuli using Mooses software (Tapp et al., 1995). The strength of these stimulus/response event pairs were assessed using Yule’s Q (Yoder & Feurer, 2000). Reactivity was examined by MeCP2 status and by complexity of cardiac signal (estimated by sample entropy) using analyses of variance techniques.

**Results:** RTT participants showed the greatest individual differences in cardiac responsivity to stimuli both in increases and decreases in heart rate. Groups differed significantly in behavioral reactivity to stimuli (df=22, p<0.01, F=6.76) but did not significantly differ in their cardiac reactivity for either increases (df=26, p<.18, F=1.84) or decreases in heart rate (df=29, p<.70, F=.35). The TYP group showed the most consistent behavioral responsivity to stimuli with DUP participants showing intermediate responsivity and RTT participants showing weakest reactivity. Cardiac signal complexity did not account for these differences in reactivity.

**Discussion:** Time lag sequential analysis can offer a parsimonious way to extract and analyze relationships of interest from large within subject data sets. In this case, the results of the sequential analysis showed differing behavioral responsivity to calibrated sensory stimuli based on MeCP2 genetic impairment. Results suggest that lag sequential analysis may offer an adjunct to other methods in dealing with the challenges of high resolution data and observable behaviors of interest. Alternative methods of analysis (e.g. cross recurrence quantification) and the flexible application of these methods to other types of behavioral (e.g. quantitative movement data) and biological (e.g. skin conductance) data streams are discussed.

**Key References:**


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Participant ID: 22

Symposium Title: Delineating Behavioural Phenotypes of Genetic Disorders Associated With Autism Spectrum Disorder

Chair(s): Lucy Wilde, Cerebra Centre for Neurodevelopmental Disorders, University of Birmingham

There is increased prevalence of autism spectrum disorder (ASD) in individuals with genetic syndromes. This symposium presents studies which examine the behavioural phenotypes of genetic syndromes associated with autism spectrum disorder (including fragile X syndrome, Smith-Magenis syndrome and Phelan-McDermid syndrome). Fine grained descriptions of behaviour and cognitive abilities associated with presentation of ASD are derived from between syndrome contrasts of behaviour and ability (including contrasts with non-syndromic autism) and through tracing the development of behaviour over time. Increased understanding of the behavioural phenotypes of genetic syndromes associated with autism have implications both for understanding the phenomenology of ASD symptomology in these syndromes and for intervention.
**Introduction:** The behavioural phenotype of Phelan-McDermid syndrome (PMS) is relatively unknown. Research has indicated atypically high levels of activity, impulsivity and autism spectrum disorder (ASD) behaviours. Divergent profiles of ASD in PMS are reported, with some studies demonstrating similarities to idiopathic ASD and others indicating an uneven profile of the triad of impairments. An evaluation of the behavioural phenotype of PMS and the prevalence and phenomenology of ASD is warranted, particularly given the putative causal involvement of the SHANK3 gene in the aetiology of PMS.

**Methods:** Carers of individuals with PMS, (N = 30; mean age = 10.55, SD = 7.08) completed questionnaires relating to impulsivity, overactivity, mood, interest and pleasure, repetitive behaviour and ASD phenomenology. These data were compared to data from matched samples of individuals with Fragile X and Down syndromes, and idiopathic ASD. In order to evaluate the profile of ASD phenomenology in PMS, two comparisons were made; first, including the total sample with PMS and second, including only those who met clinical threshold for autism on the screening measure.

**Results:** The results revealed lower mood in individuals with PMS, but no difference in impulsivity and overactivity compared to the comparison groups. Compulsive and routine driven repetitive behaviours were less common in the total sample with PMS; however, motor based stereotyped behaviours were more common. ASD phenomenology was highly prevalent, with 87% of the sample meeting criteria for ASD and 57% meeting criteria for autism. The profile of ASD phenomenology in the total sample with PMS was heterogeneous across the triad of impairments. However, the profile of those who met clinical threshold for autism was homogenous, and analogous to those with idiopathic ASD.

**Discussion:** ASD phenomenology is common within PMS. Whilst the total sample may display an atypical profile of ASD behaviour, the profile in those who meet clinical thresholds for autism is very similar to those with idiopathic ASD. These results are discussed in relation to the wider behavioural phenotype.

**References:**


**Symposia Title:** Delineating Behavioural Phenotypes of Genetic Disorders Associated With Autism Spectrum Disorder

**Chair:** Lucy Wilde

**Paper Title:** Lifespan Changes in Levels of Repetitive Behaviour in Angelman, Cornelia De Lange, and Fragile X Syndromes: An Eight-Year Follow-up

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Chris Oliver, Cerebra Centre for Neurodevelopmental Disorders, University of Birmingham

**Introduction:** The term 'repetitive behaviour' is used to describe behaviours that are repeated frequently, without variability and are inappropriate or unusual within the situation in which they occur (Turner, 1997). As with other aspects of behavioural phenotypes, there is currently a scarcity of lifespan research relating to repetitive behaviour in rare genetic syndromes. Whilst differential behavioural profiles have been identified for repetitive behaviour within some of these groups (Moss et al, 2009), these profiles have yet to be examined in a comparative longitudinal study. This study aims to describe the changing levels of repetitive behaviour across the lifespan in three genetic syndromes associated with intellectual disability.

**Methods:** Parents/carers of individuals with Angelman (AS; N=37), Cornelia de Lange (CdLS; N=45), and Fragile X syndromes (FXS; N=86), completed the Repetitive Behaviour Questionnaire (RBQ) on three occasions spanning eight years. We compared levels of repetitive behaviour in children and adults within each syndrome group and modelled latent growth curves to describe the trajectories of repetitive behaviour in these syndromes over the three measurement occasions.

**Results:** Each syndrome displayed a distinct trajectory in their levels of repetitive behaviour. In the AS group, repetitive behaviour increased in the children but remained stable in the adults. In the CdLS group repetitive behaviour was higher in the adults than the children (p<.05), however, the adults' repetitive behaviour decreased over time (p<.05), whilst the children's repetitive behaviour remained stable. In the FXS group repetitive behaviour increased in children and decreased in adults creating a significant time point by age interaction (p<.05). Latent growth curves suggest a linear trajectory in FXS and AS and a quadratic trajectory in CdLS.

**Discussion:** The presence of repetitive behaviour may vary across the lifespan in individuals with genetic syndromes. A developmental perspective should be taken when considering interventions targeted towards repetitive behaviour in these populations.

**References:**


**Symposia Title:** Delineating Behavioural Phenotypes of Genetic Disorders Associated With Autism Spectrum Disorder

**Chair:** Lucy Wilde

**Paper Title:** Features of Autism Spectrum Disorder in Smith-Magenis Syndrome: A Comparison With Idiopathic Autism Spectrum Disorder and Rubinstein-Taybi Syndrome

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**Introduction:** The profile of autism spectrum disorder (ASD) characteristics differs between different genetic neurodevelopmental syndrome groups, and in a number of syndrome groups may also differ from that seen in idiopathic ASD. Features of ASD in Smith-Magenis Syndrome (SMS) have not been studied extensively in relation to other syndrome groups.

**Methods:** Data from the Social Communication Questionnaire (SCQ) were analysed for 50 participants with SMS, alongside 50 participants with idiopathic ASD and 50 participants with Rubinstein-Taybi syndrome (RTS), matched for self-help ability and age. A second analysis was conducted for the 35 participants with SMS whose scores on the SCQ met the cut-off indicative of ASD, alongside 35 matched participants with idiopathic ASD and 35 with RTS who also met the SCQ cut-off for ASD. Group differences in subscale scores were assessed, as well as differences between the frequency with which "no" and "yes" responses were given to individual questionnaire items in the different groups. Because of the number of tests, an alpha level of 0.01 was employed. However, due to the exploratory nature of the study we also discuss where there were differences at p < 0.05.

**Results:** All data: 35 (70%) of the 50 participants with SMS, and 32 (64%) with RTS, met criteria indicative of ASD.

SMS and RTS groups scored similarly at subscale level, but a significantly greater proportion of participants in the SMS group were reported to have asked socially inappropriate questions, engaged in self-injurious behaviour, and to have a particular or best friend. At p < 0.05, participants with SMS were more likely than those with RTS to have used odd phrases repetitively, to have been interested in parts of a toy as opposed to the purpose for which it was intended, or to have been unusually interested in the sight, smell or sound of people, and were less likely to have displayed complicated repetitive body movements. As expected, subscale and total scores on the SCQ were lower (fewer ASD features) in the SMS than idiopathic ASD group, and on a number of items (especially in the communication and social interaction subscales) participants with idiopathic ASD were more likely to score in manner consistent with features of ASD. However, for items related to self-injury and the asking of socially inappropriate questions, it was the SMS group whose scores were significantly higher.

**ASD cut-off data:** Whilst there were no significant differences between the SMS and RTS groups at total score or subscale level, complicated repetitive body movements were less common in people with SMS than with RTS. As expected, the idiopathic ASD group displayed more ASD-like traits than the SMS group on the communication and reciprocal social interaction subscales. There was no significant difference on the repetitive and restrictive behaviour subscale. Individual item scores appeared to largely reflect this pattern.

**Discussion:** People with SMS display different patterns of ASD-related behaviour from those with RTS or idiopathic ASD. In people with SMS meeting criteria for ASD, behaviour may be more similar to idiopathic ASD in restricted and repetitive behaviours than in communication and social interaction. Profiles of ASD characteristics in SMS and RTS are similar at subscale level, but may differ at the level of individual behaviours.

**References:**


Introduction: Nearly all males with fragile X syndrome (FXS) are likely to demonstrate some behaviors that are characteristically observed in individuals with nonsyndromic autism spectrum disorder (ASD), with a significant proportion of males with FXS displaying symptoms frequent and severe enough to warrant a comorbid diagnosis of ASD. The behavioral similarities observed between these two phenotypes have led to the claim that similar treatment approaches (pharmacological and behavioral) will be equally beneficial in both conditions. It is possible, however, that these superficial behavioral similarities mask important differences in the underlying psychological and neurobiological mechanisms producing these behaviors in the two disorders. Understanding these mechanisms, however, requires going beyond evaluating the similarities and differences in ASD symptoms and, instead, directly comparing behavioral profiles in other domains of functioning, particularly characteristics that are frequently implicated in both phenotypes. One domain that is frequently observed to be problematic in both FXS and ASD is language development; language delay is observed in nearly all males with FXS and is often the first recognized symptom of developmental difficulty for children with nonsyndromic ASD. A comparison of the language development of children with FXS and children with nonsyndromic ASD is particularly interesting given that both disorders are characterized by difficulties effectively participating in reciprocal social interactions, a developmental pattern that can be expected to negatively influence the ways in which language is learned. In the present study, the lexical and grammatical abilities, as well as their correlates, were compared between males with FXS and males with nonsyndromic ASD.

Methods: Participants were 51 boys with FXS (M(CA) = 7.52, M(Nonverbal IQ) = 58.63) and 36 boys with nonsyndromic ASD (M(CA) = 7.43, M(Nonverbal IQ) = 65.69) who ranged from 4 to 10 years of age and had nonverbal IQs less than or equal to 85, a cut-off inclusive of essentially all males with FXS (Hessl et al., 2009). Each participant was administered measures of nonverbal cognitive ability (Leiter International Performance Scales-Revised), receptive vocabulary (Peabody Picture Vocabulary Test), expressive vocabulary (Expressive Vocabulary Test), receptive grammar (Test of the Reception of Grammar), expressive grammar (Comprehensive Assessment of Spoken Language - Sentence Completion), autism symptomatology (Autism Diagnostic Observation Schedule), and challenging behaviors (Anxiety, Depression and Mood Scale).

Results: After controlling for nonverbal cognitive ability and ASD symptom severity, between-group differences were observed for receptive (p < .001) and expressive vocabulary (p = .002), with boys with nonsyndromic ASD demonstrating weaker lexical abilities than boys with FXS. No between-group differences were observed for receptive (p = .27) and expressive grammar ability (p = .20), although both groups displayed limited variability in standardized measures of these skills. The patterns of correlations among language abilities, challenging behaviors, and ASD symptom severity differed between boys with FXS and boys with nonsyndromic ASD.

Discussion: In addition to language difficulties, individuals with FXS and individuals with nonsyndromic ASD often demonstrate some level of difficulty in participating in reciprocal social interactions. Our data suggests that, after controlling for nonverbal cognitive ability and autism symptom severity, males with FXS have larger lexicons, both receptively and expressively, than do males with nonsyndromic ASD. In addition, between-group differences were observed in the factors associated with language abilities, which suggests that different mechanisms underlie the lexical problems of the two syndromes. Clinical and theoretical implications will be discussed.

References:

The identification of sensitive clinical biomarkers that could be used for refining the early phenotype in infants with fragile X syndrome (FXS) may aid efforts to implement intervention studies. A major difficulty has been the identification of meaningful outcome targets. The availability of biomarkers in FXS would provide new and arguably more successful avenues for clinical research studies. This is particularly true for markers specific to the highly plastic early developing brain as interventions during the first years of life may have a larger and more lasting impact. Existing studies of toddlers and older children with FXS have found evidence of atypical brain growth and aberrant white matter development, as well as differences in physiological measures, such as heart rate variability and brain function as measured by ERP. This symposium will present research on infants with FXS focused on early emerging differences in brain development and pathophysiology. The first paper examines early subcortical differences in brain development in infants with FXS. Striking enlargement in the growth and size of striatal regions were observed in the infants with FXS compared to controls. The second paper describes longitudinal differences in the structural properties of white matter fiber pathways connecting cortical and subcortical brain regions in infants with FXS. Finally, we present work that finds group differences in response to social versus nonsocial stimuli on ERP measures collected from infants with FXS compared to controls and infants at high risk for autism. Additionally, those FXS infants with elevated symptoms of autism had elevated responses to stimuli on indicators of stress/anxiety (heart rate variability). These three studies suggest the potential for new and actionable biomarkers in FXS that could benefit the development of early interventions for FX.
Introduction: Previous research by our laboratory and others have shown that young children with fragile X syndrome (FXS) have enlargement of subcortical structures, including the caudate, by 2-5 years of age (Hazlett et al., 2009; Hazlett et al., 2012). However, it is currently unknown whether this altered brain trajectory can be detected in infancy. We conducted a longitudinal brain imaging study and hypothesized that infants with FXS would show altered patterns of brain growth morphology, particularly enlargement of the striatum, compared to infants later diagnosed with ASD and control infants.

Methods: Longitudinal T1- and T2-weighted imaging data were collected during natural sleep at 6, 12, and 24 months in N=20 infants with FXS, N=50 infants who were later diagnosed with autism spectrum disorder (ASD), and N=131 age-matched control infants. A total of 38 useable scans were acquired in the infants with FXS between 6-24 months, 123 total scans were acquired in the ASD group, and 278 total scans were acquired in the control group. T1 and T2 images underwent distortion correction, mutual registration, CSF/tissue segmentation, and multi-atlas subcortical segmentation was used to generate volumes for the left and right caudate, putamen, globus pallidus, thalamus, amygdala, and hippocampus. A repeated-measures mixed effects model was used to test age trends, interactions and main effects of group, while controlling for differences in age, sex, and intracranial volume.

Results: Preliminary analyses revealed that the FXS group had significantly larger volumes of striatal structures (all p<.005; controlling for age, sex, and intracranial volume) compared to the other groups by 12 months of age, including the bilateral caudate (18% larger than the control group; 15% larger than ASD), globus pallidus (18% larger than controls; 16% larger than ASD), and putamen (9% larger than controls; 7% larger than ASD). At 24 months of age, the FXS group continued to have significantly larger striatal volumes (p<.005), including the bilateral caudate (20% larger than the control group; 20% larger than ASD), globus pallidus (28% larger than controls; 28% larger than ASD), and putamen (16% larger than controls; 16% larger than ASD). The thalamus was also significantly enlarged in the FXS group compared to the other groups (p<.01) at both 12 months (6% larger than controls; 3% larger than ASD) and 24 months (16% larger than controls; 13% larger than ASD). The amygdala was significantly smaller in the FXS group at 12 months compared to the ASD group (6% smaller; p<.05), but no differences were found at 24 months. There were no significant differences between groups in hippocampal volume.

Discussion: These results indicate that infants with FXS have a striking enlargement of the striatum compared to both infants who develop ASD and age-matched controls. These results are consistent with earlier reports found in toddlers (Hazlett et al., 2009; Hazlett et al., 2012), which suggests that the increased size and growth of striatal regions (including the caudate, putamen, and globus pallidus) is an early neural abnormality in FXS that can be detected from infancy through early childhood. In addition, while FXS shares behavioral characteristics with idiopathic autism, it appears that longitudinal brain imaging can delineate distinct brain trajectories starting in early life. We will examine the associations between the growth of striatal brain regions and behavioral symptoms that may underlie these regions, including motor stereotypies and anxiety measures, to evaluate whether aberrant growth of the striatum could serve as an early biomarker for clinical symptoms that arise in children with FXS.

References:


Symposia Title: Neurobiological and Neurophysiological Characteristics of Infants With Fragile X Syndrome

Chair: Heather C. Hazlett

Paper Title: A Longitudinal Diffusion Tensor Imaging Study of Infants With Fragile X Syndrome

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Introduction: There is ample evidence that atypical brain growth is associated with fragile X syndrome (FXS) early in life (Hazlett et al. 2012; Hoeft et al. 2010). While studies of older individuals has also implicated white matter neural circuitry in FXS (Barnea-Goraly et al. 2003; Hashimoto et al. 2011; Villalon-Reina et al., 2013), nothing is yet known about the role of structural connectivity in FXS during infancy. Our aim was to characterize white matter development using diffusion tensor imaging (DTI) in a longitudinal sample of infants with and without FXS.

Methods: This sample included 10 infants with FXS (30% female) and 61 typically developing infants (37% female). Participating babies provided brain imaging data during natural sleep at 6 and/or 12 months of age, with most infants providing data at both times. For this study, we elected to focus on three divisions of the corpus callosum (genu, body, and splenium), anterior and posterior corona radiata, arcuate fasciculi, uncinate fasciculi, and superior cerebellar peduncles (SCP). Data processing and fiber tractography employed a previously described approach (Verde et al., 2013; Wolff et al. 2012). Microstructural properties of defined fiber pathways were characterized by fractional anisotropy (FA), an index of magnitude of directional diffusion, as well as axial and radial diffusivities, which reflect diffusion along and orthogonal to the fiber pathway, respectively. Longitudinal imaging data were analyzed using repeated measures LMM.

Results: There were no differences between groups by age (Time 1: p = 0.941; Time 2: p = 0.70) or sex (p = 0.51). As expected, groups differed significantly in cognitive development as indexed by the MSEL, p < .001. The effect of Time was significant for all fiber pathways (p < .000). There were group effects for the Genu (F = 14.3, p < .000) and Body (F = 6.3, p = .016) of the corpus callosum. Group effects were also identified for the anterior corona radiata (Left: F = 6.5, p < .013; Right: F = 8.1, p = .006), the right SCP (F = 5.4, p = .024), and the uncinate fasciculi (Left: F = 6.7, p < .011; Right: F = 6.5, p = .013). The splenium, posterior corona radiata, left SCP, arcuate fasciculi did not differ significantly between groups, and there were no significant effects for the interaction of Group X Time.

To elucidate FA findings, we next examined radial and axial diffusivities. We found that radial, but not axial diffusivity, differed significantly between groups for most pathways, with group differences highly pronounced in the right SCP, uncinate fasciculi, and genu. For these latter two pathways, there was an additional effect for Group X Time. Overall, babies with FXS were characterized by lower FA and higher RD. Though sample size was insufficient to examine sex differences in FXS, visual analysis indicated no qualitative differences between male and female babies.

Discussion: Early development of neural circuitry is rapid in pace and includes both exuberant growth (myelination, arborization) and regressive events (axon elimination). The present results suggest that these developmental processes may be significantly altered during infancy in FXS, implicating either decreased fiber density/organization, less myelination, or both. Overall these findings are consistent with previous work and add further evidence that atypical brain growth in FXS may be concomitant with a less developed and arguably less efficient set of structural connections between brain regions. Limitations include potential sources of error impacting imaging data, such as partial volume effects. Next steps include expanding the sample and following children to age 2 to establish early brain-behavior relationships, as well as adding comparison groups including IQ-matched controls and babies who develop idiopathic autism.
**Symposia Title:** Neurobiological and Neurophysiological Characteristics of Infants With Fragile X Syndrome

**Chair:** Heather C. Hazlett

**Paper Title:** Biobehavioral Correlates of Autism Spectrum Disorder in Infants With Fragile X Syndrome

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**Introduction:** Young males with fragile X syndrome (FXS) are at high risk for autism spectrum disorders (ASD) with up to 70% meeting diagnostic criteria. While early identification efforts in idiopathic non-fragile X ASD have accelerated given the known benefits of early treatment, relatively little work exists focused on early detection of autism in FXS. Identification of ASD in very young children, however, is fraught with challenges given the complexity of identifying ASD amidst the rapid developmental changes in social behavior, cognition and language that occur in early childhood. Consequently, consideration of prodromal and symptom level factors during infancy has increased as has recognition of the role of biomarkers in identifying putative mechanistic factors. The overarching aim of this presentation is to identify the relationship of multiple biomarkers to ASD features in infants with FXS contrasted to typically developing infants (TD) and infants with older siblings diagnosed with ASD (SIBS). We will present data reflecting neural correlates (event-related potentials) of face processing and baseline autonomic measures (heart activity) in relation to ASD features.

**Methods:** Across both ongoing studies, 54 infants 12 months of age (17 FXS, 17 SIBS, 20 TD) participated. Autism symptoms were assessed using the Autism Observation Scale for Infants, and ASD diagnoses were determined using the Autism Diagnostic Observation Schedule 2. Event related potentials were measured in response to images of the child's own mother (familiar social), favorite toy (familiar object) contrasted to an unfamiliar woman, and unfamiliar toy (500 ms duration). High-density EEG's were recorded using a 128-channel net. Grand averages and peak amplitudes were calculated to capture the N290. The Nc was calculated from 350 to 700 ms post-stimulus onset. Heart activity was also collected during a passive viewing task as part of the larger battery in a subset of participants (9 FXS, 15 SIBS, 12 TD). Dependent measures included inter-beat interval, respiratory sinus arrhythmia, and proportion of time in heart-defined sustained attention, a period of decelerative heart rate that is associated with greater stimulus engagement in typically developing infants (e.g. Richards & Casey, 1991). We anticipate increased samples for both studies for the presentation.

**Results:** Results from the ERP study indicated a main effect for trial (F(1,88)=4.96; p<.01) with a larger amplitude for faces than toys for all groups for the N290. Results also indicated a marginal interaction effect (F(2,33)=2.79; p<.08) suggesting a larger effect for the toy than face stimuli for the FXS and SIBS groups. Also, evidence suggested that infants with FXS and elevated ASD symptoms showed a larger Nc to the stranger than to their mother that was not observed in the TD, SIBS and the FXS group with a low number of ASD symptoms. Preliminary heart activity data indicated that groups did not differ in inter-beat interval, respiratory sinus arrhythmia, and qualities of heart-defined sustained attention (Kruskal-Wallis p>.10). However, across groups, higher AOSI scores were associated with greater proportion of time in heart-defined sustained attention (r=.36, p=.03) and increased behavioral looking time (r=.41, p=.02).

**Discussion:** The larger P400 in the SIBS and FXS groups may reflect an object-based preference for processing. Also, the heightened Nc response to the stranger in the FXS group with elevated ASD symptoms could signal a relationship of ASD to neural components associated with attention and cognitive processing. Heart activity data indicate that increased heart defined sustained attention predicted elevated ASD symptoms across groups. These preliminary results indicate that biomarkers are sensitive to detect early markers of risk for ASD in vulnerable populations.

**References:**

Atypical social functioning is evident in many neurodevelopmental disorders. Understanding factors which might underpin such atypicalities may facilitate targeted intervention. In this symposium studies are presented which use diverse methods to assess factors potentially underpinning atypical social behaviour in a range of neurodevelopmental disorders (including idiopathic autism, fragile X syndrome, Tuberous Sclerosis Complex and Rubinstein-Taybi syndrome). Eye tracking and ERP methods are used to examine social cognitive processes including attention and response to social stimuli. Direct assessment of social cognitive abilities is described and related to executive functions. Finally, direct observation of social behaviour is used to examine both social anxiety and social motivation as factors potentially underpinning reports of social atypicality in neurodevelopmental disorders.
**Symposia Title:** Assessing Factors Underpinning Atypical Social Functioning in Neurodevelopmental Disorders

**Chair:** Chris Oliver

**Paper Title:** Salience Affects Attention to Social Stimuli in Children With Tuberous Sclerosis Complex

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**Introduction:**
Tuberous Sclerosis Complex (TSC), is associated with increased rates of Autism Spectrum Disorder (ASD). Prevalence estimates vary but commonly cited rates range from 24% to 60%. A number of studies have indicated that processing of social stimuli may be atypical in individuals with ASD, with reduced attention to social, compared to non-social, stimuli reported. It has been suggested that reduced allocation of attention to social stimuli may result in reduced engagement in social interactions and subsequent difficulties with social interaction. Given increased prevalence of ASD in TSC atypical social processing may be evident in this disorder. Past research has found atypicalities in face processing in children with TSC (slower face processing and lack of expected hemispheric differences) using event-related potentials. To further explore whether attention to social stimuli is atypical in TSC the current study used eye tracking methodology to examine spontaneous attention allocation to social versus non-social stimuli in children with TSC.

**Methods:**
Eye tracking methodology was used with seventeen children with TSC (10 male, 7 female, mean chronological age 8.2 years) and a contrast group of 25 typically developing (TD) children (15 male, 10 female, mean chronological age 5 years). Inclusion criteria for typically developing children included scoring below the cut off score for ASD on the Social Communication Questionnaire and not having a diagnosis of a developmental disorder. No significant difference was found between estimates of developmental age derived for the samples (using chronological age as a proxy for developmental age for the TD sample and using standardised assessments e.g. British Ability Scales, Mullen Scales of Early Learning, for the TSC sample). Children viewed paired video clips presented side by side, with one social video (showing a person) and one non-social video (showing objects). Half of these paired videos were directed (subject facing towards the camera) and half were non-directed (subject not facing towards the camera).

**Results:**
Both dwell time (percentage time looking at social and non-social videos) and time to first fixation to social and non-social videos were analysed, comparing 'preference' for social versus non-social videos. Analyses employed mixed ANOVAs. No differences were found between groups in dwell time. However, for time to first fixation an interaction suggests that when videos were directed preference for social videos (over non-social videos) did not differ between groups but when they were non-directed preference for social videos became significantly weaker in children with TSC in comparison to typically developing children.

**Discussion:**
Findings suggest potential atypicalities in the allocation of attention to social and non-social stimuli when stimuli are less salient (i.e. not directed at the individual), with children with TSC showing reduced social preference for less salient stimuli. Give the importance of attending to social stimuli for social development, this reduced attention to social stimuli which are less salient may provide insight into a potential pathway to the social impairments observed in TSC.

**References:**


**Symposia Title:** Assessing Factors Underpinning Atypical Social Functioning in Neurodevelopmental Disorders

**Chair:** Chris Oliver

**Paper Title:** Incidental Memory for Faces: ERP Index of Increasing Social Interest Following SENSE Theatre Treatment in Children With Autism

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**Introduction:** Ability to remember and recognize faces is important for successful social functioning (Schultz et al., 2005). Children and adults with autism spectrum disorders (ASD) are often reported to have difficulty with memory for faces (Boucher et al., 1998; Dawson et al., 2002; Klin et al., 1999). Recently we used event-related potentials (ERPs) to demonstrate that faces are a highly salient stimulus that can elicit a memory trace (parietal P300 response) even during passive viewing in typical children, but not in children with ASD (Key & Corbett, 2014). The purpose of the current study was to investigate whether a treatment program targeting social skills (SENSE Theatre) in children with ASD may improve their incidental memory for novel faces.

**Methods:** Visual ERPs were recorded in 34 8-14-year-old children with ASD who comprised a treatment (n=17) and wait-list control (n=17) groups. All participants completed two test sessions (before and after SENSE Theatre; 3 months apart) in which they viewed a sequence of color photographs depicting unfamiliar smiling young adults or front views of houses. One face and one house were randomly selected and repeated 50 times throughout the test session while the rest of the pictures were shown only once (50 trials). Social skills were assessed using Social Communication Questionnaire (SCQ), Social Responsiveness Scale (SRS), and Adaptive Behavior Assessment System (ABAS) measures. Ability to remember faces was also evaluated using NEPSY Memory for Faces test.

**Results:** Repeated measures analysis of variance with Group (treatment, wait-list) as the between-subject factor and session (pre, post) x stimulus (face, house) x repetition condition (single, repeated) revealed no group differences in the mean amplitude of the early ERP responses reflecting comparable perceptual processing as both groups elicited larger N170 responses to faces than houses. Similarly, there were no group differences in the parietal P300 response to repeated vs. novel faces or houses during the pre-treatment session. In the absence of treatment, ERP responses indexing incidental memory remained stable over the 3-month test-retest interval. Conversely, the treatment group demonstrated the increased parietal response to repeated faces (similar to the ERP responses observed previously in typical children (Key & Corbett, 2014) and the old/new effect reported in studies requiring active memorization). There were no pre- vs. post-treatment differences in responses to houses. All participants demonstrated reduction in the parietal response to the repeated image, consistent with previous fMRI reports of decreased activation of the novelty-detecting network in response to familiarized nonsocial stimuli (Jessen et al., 2002). The strength of the memory trace for faces was not related to age, IQ or ADOS scores.

**Discussion:** Reduced salience of social stimuli in children with ASD is associated with less extensive post-perceptual face processing as reflected by poor incidental memory during passive viewing. These results indicate that completion of the SENSE Theatre treatment was associated with increased salience of social information, reflected in faces receiving sufficiently extensive processing to form a memory trace beyond perceptual familiarity even in the absence of explicit instructions directing attention to the stimuli.

**References:**

Introduction: Associations between executive function (EF) and theory of mind (ToM) have been established in the typically developing literature. In particular, EF development may be prerequisite to fully fledged ToM, which depends on holding multiple perspectives in mind, while inhibiting previously held perspectives. Exploring this association in intellectual disability is important for understanding how individuals navigate the social world. For example, individuals with Rubinstein-Taybi syndrome (RTS) are described as having reduced stranger awareness and research has implicated deficits in ToM and EF. To date, specific associations between components of EF and ToM have not been explored in this group.

Methods: Twenty-four individuals with RTS (mean age: 20.89; range = 6.75 - 44.42) were visited at home. Participants completed a measure of general cognitive ability (The Mullens Scales of Early Learning or Wechsler Abbreviated Scale of Intelligence) and scaled batteries of ToM and EF tasks (inhibition, working memory and switching). These batteries were previously adapted for individuals with a moderate to severe level of intellectual disability. Parents completed the Behaviour Rating Inventory of Executive Function - Preschooler (The BRIEF-P).

Results: Each participant was assigned a ToM developmental age based on their performance. A strong positive association was found between the ability to inhibit responses on the experimental battery and greater ToM developmental age (R = .70; p < .001). This pattern was replicated in the parent ratings of inhibition; better inhibition on the BRIEF-P was associated with higher ToM developmental age (R = -.41; p = .034). A greater working memory capacity was associated with ToM task performance on the experimental battery (R = .81; p < .001) and questionnaire measure (-.50; p = .011). Partial correlations were conducted to control for mental age. The associations between ToM and the inhibition tasks and parental ratings of inhibition remained significant (R = .43; p = .020 & R = -.40; p = .042); however, working memory was no longer associated with ToM.

Discussion: This study is the first to explore associations between executive functions and theory of mind ability in individuals with RTS. The results suggest that difficulties with inhibition could account for poor performance on ToM tasks in this group. Further research should explore the direction of this association. These results may be important for understanding behavioural characteristics associated with RTS that put individuals at risk of social exploitation. When working with individuals with RTS around stranger awareness it may be important to help individuals with RTS develop their ability to inhibit responses. The importance of understanding EF and ToM in the wider intellectual disability population will be discussed.

References:
Introduction: Increased social anxiety, alongside a willingness to interact, have been reported to describe the social impairments in individuals with Fragile X (FXS) and Cornelia de Lange (CdLS) syndrome, whereas typical social interest and intact social skills are reported in individuals with Rubinstein-Taybi syndrome (RTS). Understanding whether particular syndrome groups are at risk for heightened social anxiety, and understanding the situations in which social anxiety may be induced is important for intervention planning. In this study, the effects of adult familiarity and type of social interaction on social anxiety and social motivation were investigated.

Methods: Individuals with FXS (n = 20), RTS (n = 20), CdLS (n = 20) and Down syndrome (DS; n = 20) participated in four social tasks, each with a familiar and unfamiliar adult. The four social tasks differed in the level of the social demand and consisted of no social interaction, voluntary social interaction, required social interaction and a performance. Social anxiety and motivation was assessed using the Social Anxiety and Motivation Rating Scale, developed for this study.

Results: To assess social anxiety, a 4 (condition) x 2 (familiarity) x 4 (syndrome) mixed ANOVA revealed a three-way interaction (p = .003). Further analyses revealed that whilst participants with FXS and RTS exhibited high levels of social anxiety during all social interactions with both familiar and unfamiliar adults, participants with CdLS showed heightened social anxiety during the voluntary social interaction, and when interacting with an unfamiliar versus familiar adult. A three-way interaction was also revealed for socially motivated initiation of interaction (p = .014). Further analyses revealed that participants with FXS and RTS initiated more interactions with unfamiliar versus familiar adults, whereas participants with CdLS initiated fewer interactions with unfamiliar adults compared to other syndrome groups, during the voluntary social interaction condition.

Discussion: These results indicate that social anxiety is high but consistent across interactions with familiar and unfamiliar adults in individuals with FXS and RTS. However, in CdLS, social anxiety is more likely to be influenced by the nature of the social situation and the familiarity of the interacting adult. It is suggested that the heightened social anxiety in individuals with CdLS during the voluntary social interaction may stem from the ambiguity of the social norms and expectations during this condition. Furthermore, the results show that whilst social anxiety and social motivation are related constructs, they are not dependent on one another. This study has highlighted genetic syndrome groups most at risk for heightened social anxiety and investigated the social situations that are most likely to elicit social anxiety in children and adults with FXS, CdLS and RTS.

References:


Symposium Title: Executive Function, Academics, and Functional Performance in Down Syndrome

Chair(s): Deborah Fidler, Colorado State University

From early childhood, individuals with Down syndrome have a high probability of demonstrating a distinct cognitive profile including deficits in verbal processing, working memory, and goal-directed behavior (for a review see Daunhauer & Fidler, 2011). Although Down syndrome is the most common neurogenetic syndrome associated with intellectual disability (Parker et al., 2010), significant gaps remain in our characterization of cognitive phenotype associated with Down syndrome. Recent evidence suggests that individuals with Down syndrome demonstrate impairments in executive function (Kogan et al., 2009; Lee et al., 2011; Rowe, Lavender, & Turk, 2006). Furthermore, in a review of the DS neuroanatomical phenotype, Nadel described specific reductions in the size of the prefrontal lobes (2003), an area of the brain associated with executive function (Tau & Peterson, 2010). Yet, despite converging neuroanatomical and behavioral evidence, a paucity of comprehensive work has been conducted on the development of executive function in young children with Down syndrome. In this symposium, our team reviews its most recent findings regarding executive function in school-age children with Down syndrome. Specifically, we will examine: · approaches to measuring executive function in early development for students with Down syndrome and an MA-matched comparison group; · executive function as a predictor of math achievement in students with Down syndrome; and · moderating effects of short-term memory on academic performance in Down syndrome; · the relationship between functional performance and executive function in students.
Executive function (EF) is an area of relative developmental weakness in adolescents and adults with Down syndrome (DS; Lanfranchi et al., 2010; Rowe et al., 2004). There is also evidence that specific aspects of EF, such as working memory and planning, may be more impaired in DS than other EF component skills (Fidler et al., 2014; Kasari & Freeman, 2001). To date there have been no comprehensive studies of lab-based EF skills in young students with DS. Further complicating this area of inquiry is the lack of agreement on how to measure EF in early development, such as using lab tasks that allow for precise measurement of discrete skills or using informant reports regarding young children's EF performance in everyday contexts such as school and home life (Gioia et al., 2002). Extant research on preschoolers through adolescents with clinical diagnoses related to attention, learning, and behavior have found little association between performance on EF lab measures and informant-reported EF performance (Mahone & Hoffman, 2007; McAuley et al., 2010). It is not known how performance in these two approaches to EF measurement is similar or different in young students with DS. Given that informant-reported EF skills predict school function better than IQ in this population (Daunhauer et al., 2014); and interventions that target goal-directed behavior have been less effective for young children with DS when compared to children with other types of intellectual disability (e.g., Yoder et al., 2014), characterizing the profile of EF lab task performance in younger children with DS and its associations to everyday life will be critical to inform targeted intervention for this population.

Participants were students with DS (n=40, mean MA= 49.53, SD= 7.88) and those with typical development (TD; n=37, mean MA= 49.86, SD= 5.90) matched for nonverbal mental age using the Leiter-R (Roid & Miller, 1997). All participants were assessed with a battery of laboratory tasks that reflect the primary EF domains (working memory- modified Bear and Dragon, inhibition-Snack Delay, and cognitive flexibility- Dimensional Change Card Sort; e.g. Carlson, 2005; Zelazo & Jacques, 1996). Additionally, executive function in everyday life was measured by parent report using the Behavior Rating Inventory of Executive Function- Preschool Version (BRIEF-P; Gioia, Espy, & Isquith, 2003).

Results indicated that the DS group demonstrated significantly greater deficits in EF lab task performance as measured by a composite of the laboratory tasks (F (1,64) = 8.90, p = .004, partial eta squared=.12) and the BRIEF-P (F (1,64) = 27.64, < .001, partial eta squared=.30) Post-hoc analyses examining performance between group for specific lab tasks indicated that the DS group demonstrated significantly more difficulty on the working memory task (t (66) = -2.52, = .014, d = -.67), but performed similarly to the TD group on the inhibition and cognitive flexibility tasks with Bonferonni corrections. Furthermore, when examining the associations between lab tasks and domains of the BRIEF-P, medium to large associations were found with the lab task composite score and the working memory lab task and the following domains of the BRIEF-P: working memory, planning, and inhibition (r's range .34 to .47, p < .005).

These findings add to the growing literature on EF and EF measurement in early development for both children with DS and those with TD. The lab performance provide evidence for early working memory impairments in young students DS. The associations between lab tasks and the informant-reported BRIEF-P suggest both measurement approaches may contribute unique and shared variance in the comprehensive measurement of EF. Implications for intervention planning and future research in DS will be discussed.
Symposia Title: Executive Function, Academics, and Functional Performance in Down Syndrome

Chair: Deborah Fidler

Paper Title: Executive Function and Math Achievement in Students With Down Syndrome

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Students with Down syndrome show deficits in mathematical reasoning when compared to developmentally matched peers (Belacchi et al., 2014; Sella et al., 2013), suggesting a specific area of challenge beyond those delays anticipated when accounting for overall developmental status. To better understand the nature of this specific area of deficit, it may be useful to examine the cognitive underpinnings of mathematical reasoning difficulties, particularly in the area of executive function (EF). EF has been shown to be strongly related to and predictive of mathematical achievement in young typically developing students (Fitzpatrick et al., 2014; Miller et al., 2014; Verdine et al., 2014). Given that students with DS present with a specific profile in the area of EF and goal-directed behavior (Lee et al., 2011; Daunhauer et al., 2014; Fidler et al., 2014), understanding the associations between EF and math achievement in DS may help shape instructional and intervention approaches to improve educational outcomes in this population.

Participants were 29 students with DS (mean CA = 80.03 mos; SD = 12.92) and 31 nonverbal mental-age matched typically developing children who were chronologically younger (mean CA = 39.77 mos; SD = 5.33). Groups were matched for nonverbal mental age using the Leiter-R (Roid & Miller, 1997; DS mean age equivalent = 46.79 mos (SD= 8.93); TD = 48.26 (SD = 5.03). Child participants completed the Woodcock-Johnson III-Tests of Achievement Applied Problems domain to assess math achievement; Woodcock, McGrew, & Mather, 2001, 2007) and a laboratory based battery of EF that included assessments of working memory (modified Bear/Dragon; Carlson, 2005; Flynn, 2007; Garon, Bryson, & Smith, 2008; Kochanska, Murray, Jacques, Koenig, & Vandegeest, 1996; Murray & Kochanska, 2002), planning (generativity; Fidler et al., 2014; Rutherford & Rogers, 2003), shifting (DCCS; Zelazo, & Jacques, 1996; Zelazo, Frye, & Rapus, 1996, Zelazo, 2006), and inhibition (snack delay; Carlson 2005; Carlson, Mandell, & Williams, 2004; Kochanska, Murray, & Harlan, 2000).

Diverging patterns of association were observed when examining EF performance and math achievement (as measured by WJ-III Applied Problems) across the two groups. Pearson correlations demonstrated a significant association between working memory, planning, and inhibition with Applied Problems performance in the DS group (r’s between .36 and .55, p’s < .05). When entered into a multiple regression, working memory performance emerged as a statistically significant predictor of Applied Problems in the DS group. In contrast, working memory and shifting were strongly associated with Applied Problems performance in the TD group (r’s between .45 and .47, p’s < .05). When entered into a multiple regression, both working memory and shifting were significant predictors of Applied Problems performance in TD students.

These findings add to the growing literature on the importance of EF in early development for both TD children and children with DD. In particular, these findings also support the evidence for the importance of early “cool EF” (working memory and planning) skills in DS, and the critical role that they may play for both developmental and educational outcomes in this population. Implications for education and intervention planning in DS will be discussed.
Letter-word identification (LW-ID) is an area of relative strength that has been repeatedly identified within the Down syndrome (DS) educational profile (Abbeduto, Warren & Conners, 2007; Beaudreau, 2002; Fidler, Most, & Guiberson, 2005). Older children and young adults with DS perform at or above their overall developmental status on LW-ID tasks and perform better than mental-age (MA) matched typically developing (TD) peers (Beaudreau, 2002; Fidler et al., 2005). Strengths in this area have been linked to visual processing abilities observed in the behavioral phenotype (Beaudreau, 2002; Fidler, 2005, Fidler et al., 2005). Verbal abilities are a demonstrated skill recruited in word identification and reading abilities in DS and typical development (Jarrold, Thorn, & Stephens, 2011). Although visual processing abilities have been evaluated in DS, the specific effect of visual and verbal short-term memory remains under-evaluated. We examined the effects of short-term memory (STM) on LW-ID in children with DS compared to typical development.

In this study, we tested an academic-performance profile in children with DS (N=29; Mean MA=46 months; Mean CA=78) in comparison to mental age-matched TD children (N=25; Mean MA=47 months; Mean CA=39 months) and evaluated moderating effects of verbal and visual short-term memory (STM) on LW-ID performance. Participants were administered the Leiter-R (Roid & Miller, 1997) to obtain a brief nonverbal IQ composite. Academic performance was measured using the four subdomains of the Woodcock-Johnson III-Tests of Achievement (i.e. applied problems, quantitative concepts, letter-word identification (LW-ID), and picture vocabulary; Woodcock, McGrew, & Mather, 2001, 2007). Additionally, two subscales of the Kaufman Assessment Battery for Children (KABC; Kaufman & Kaufman, 1983) were administered measures of verbal short-term memory (Number Recall) and visual short-term memory (Hand Movement).

Multiple analysis of variance analyses indicate a significant group difference in academic performance ($F(1, 53)=20.00, p<.001$) with a large effect size ($\eta^2=.63$). Univariate ANOVAS indicate that groups differed significantly on Letter-Word ID, but no other subscales ($F(1, 52) = 23.60, p<.001$). Parameter estimates indicate that participant in the DS group performed significantly better than participants in the TD group on LW-ID (b=57.24; p<.001). Further analyses evaluated contributions of processing abilities to this effect. Moderation analyses were conducted to determine whether performance of each group on LW-ID differed as a function of verbal STM or visual STM. In the first moderation analysis, effects of verbal short-term memory on LW-ID were examined. Results indicated that verbal STM was a significant moderator of group performance on LW-ID. There was a significant predicted difference in the effect of verbal STM on LW-ID for TD compared to the DS group (b=-2.83; p<.05), such that the effect was significantly less for children in the TD group. In the second moderation analysis, the effect of visual STM on LW-ID was examined. Results indicate that visual STM was also a significant moderator of group performance on LW-ID. There was a significant difference in the effect of visual STM on LW-ID for TD compared to DS such that there was significantly less of an effect for TD (b=-2.33; p<.05).

Results indicate that children with DS show LW-ID capabilities at a level significantly greater than their overall developmental status would predict, and greater than their MA-matched peers. Results of moderation analyses indicate that both verbal and visual STM abilities have a different effect for young children with DS compared to TD children. These findings have implications for the emergence of these skills in the DS behavioral phenotype. Given these findings, children with DS may recruit different skills during LW-ID processing than TD children as a compensatory mechanism for areas of challenge within the DS phenotype, such as working memory.
Down syndrome (DS) is the most common neurogenetic syndrome associated with intellectual disability (Parker et al., 2010). Children with DS are predisposed to areas of relative developmental strength and challenge, but it is unclear whether this phenotypic profile affects functional performance. Functional performance is the engagement in activities universal to all children—such as self-care, mobility, and social interaction. While much research has focused on implications of a behavioral phenotype for individuals with DS, there is a paucity of research examining whether this phenotypic profile affects functional performance (see Daunhauer & Fidler, 2011; Daunhauer, 2011 for a review). Identifying patterns and predictors of functional performance in DS is critical as it is a foundation for optimal outcomes for the child, their family, and community. This study examined the cognitive predictors of functional performance including IQ and executive function.

Students with DS (n=31, mean MA = 51.42 months; SD = 11.01; mean CA = 89.29 months; SD=14.96) and students with mixed developmental disabilities matched for mental and chronological age (DD; n=20, mean MA =52.40 months; SD = 12.45, mean CA = 86.25 months; SD= 17.58) participated in this study. Mental age was assessed using the Brief IQ from the Leiter International Performance Scale-Revised (Leiter-R; Roid & Miller, 1997). Functional performance was measured by parent report using the Pediatric Evaluation of Disability Inventory (PEDI; Haley et al., 1992) and executive function was measured by parent report using the Behavior Rating Inventory of Executive Function- Preschool Version (BRIEF-P; Gioia, Espy, & Isquith, 2003).

Overall, students with DS did not significantly differ from students with DD on the PEDI total scores (F (6, 44) = 2.16, p = .07, partial eta squared = .23). However, for students with DS significant within-group differences were demonstrated across the functional skill and caregiver assistance domains of the PEDI (F (2, 60) = 48.37, p< .001, partial eta squared = .62 and F (2, 48.8) = 15.16 p < .001, partial eta squared = .34 respectively). Post-hoc analysis showed that students with DS reported higher levels of functional skills on mobility than self-care (t (31) = 8.38, p < .001) and social function (t (31) = 9.87, p < .001). Students with DS also reported higher levels of caregiver assistance for social function skills than mobility (t (31) = -4.62, p < .001) and higher levels of caregiver assistance for self-care skills than mobility (t (31) = -3.77, p < .001).

A multiple linear regression indicated differing significant predictors for PEDI performance by group. Executive functioning was the only significant variable to predict PEDI performance in the DS group (Beta = -.47, p = .008) while in the DD group the Brief-IQ composite was the only significant variable to predict PEDI performance (Beta = .70, p = .001).

Results indicate that individuals with Down syndrome show a distinct profile of functional performance with relative strengths in mobility and relative challenges in self-care and social function. In addition, executive function may have a critical role in the development of functional performance for DS. These findings have implications for future targeted intervention for students with DS to promote the optimal functional performance outcomes.
The purpose of this symposium is to present findings from three studies focused on interventions for students with disabilities including autism spectrum disorders (ASD) and one study comparing the verbal behavior of children with ASD to that of children with language delay on naturalistic measures of spoken language. The first study examined the effectiveness of student-made social stories on social initiations of students with ASD. Findings suggested that student-made social stories were fairly effective to increase social initiation skills across three participants during intervention and maintenance phases. However, target skills were not generalized to different people. Social validity data collected from parents and teachers also indicated that student-made social stories were effective to promote social initiation skills. The second study compared 53 students with language delays and 17 students with ASD on four measures of naturalistic language. Findings suggested that the total number of words on the naturalistic language ample was a significant predictor of ASD. The third study examined the effectiveness of teachers' use of the system of least prompts and contingent imitation on the unprompted, generalized and novel pretend play behaviors of four children with disabilities and their Individualized Education Program goals. Results indicated the teachers' use of the intervention package was functionally related to increase in the children's frequency and diversity of pretend play and related behaviors. The discussion will address four key questions: a) what is the effectiveness of student-made social stories on social communications of students with ASD? b) What is the effectiveness of teachers' use of the instructional package (i.e., the system of least prompts and contingent imitation) on the unprompted, generalized and novel pretend play behaviors of children with disabilities and their Individualized Education Program goals? and c) What is the predictor of ASD based on the naturalistic language sample?
Symposia Title: Social Communication Skills of Students With Disabilities Including Autism Spectrum Disorders

Chair: Cathy Qi

Paper Title: Using Individualized Student-Made Social Stories to Increase Social Initiations for Students With Autism Spectrum Disorder

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Introduction: Students with autism spectrum disorders (ASD) demonstrate deficits in areas of communication, socialization, as well as interests and activities (American Psychiatric Association, 2000). Social story intervention has been used to improve social behaviors, as well as decreasing behavior challenges (Quilty, 2007). Language experience approach (LEA) is a method incorporating an individual reader’s language experiences into an intervention (Ward, 2005). Few studies have incorporate LEA into behavior intervention for students with ASD. The purpose of this study is to incorporate children’s language experience to create individualized social stories on increasing social initiations for students with ASD.

Methods: A multiple baseline design across participants was used in this study. A randomization to assign participants to different intervention starting point was conducted. Three students with ASD (two males and one female) aged 6 to 12 participated in the study. Social stories were developed based on Gray's (2000) guidelines and each child's language experience and created by the students themselves along with the teachers’ assistance. After the stories were written, each teacher read the story once to each participant prior to the observation sessions. Each teacher asked comprehension questions after reading the stories. Data was collected 3 times a session across baseline, intervention, maintenance, and generalization phases in a local art center serving students with ASD on weekends. A total of 30 sessions of data were collected for each participant. The inter-observer reliability was 96% across 20% of all sessions. The procedural fidelity was 100% for all participants.

Results: Visual analysis showed that student-made social stories were effective across three participants during intervention and maintenance phases. In addition, percentage of non-overlapping data (PND; Scruggs, Mastropieri, & Casto, 1987), percentage of data points exceeding a Median (PEM; Ma, 2006), and pairwise data overlap squared (PDO2; Parker & Vannest, 2007) also were utilized to evaluate the effectiveness of the intervention. The overall mean of PND scores across participants and phases was .40 (SD=.36, Range =0-1). The mean of PEM scores across participants and phases was .78 (SD=.28, Range =.33-1). The mean of PDO2 across participants and phases was .83 (SD=.19, Range=.54-1). The PEM and PDO2 results suggested that student-made social stories were fairly effective to increase social initiation skills across three participants during intervention and maintenance phases. However, PND scores showed that student-made social stories intervention was questionable. Target skills were not generalized to different people. Social validity data collected from parents and teachers indicated that student-made social stories were effective to promote social initiation skills.

Discussions: Findings from this study suggested that student-made social stories were effective in increasing social initiations with ASD and maintaining over time. The specific social stories created by the participants serve as a natural reinforce, which provides strong motivation. However, students did not generalize the target skills to different people. Future studies should plan generalization probes by teaching generalizing skills. The limitation of the study was that the intervention was conducted in an art center in which the instruction was given in a relaxed and easy manner, thus findings cannot be generalized to other settings. Future research should apply this strategy in more naturalistic settings.

References:


Introduction: Autism spectrum disorder (ASD) is a developmental disorder that is characterized by deficits in social communication and restricted or repetitive interests (APA, 2013). Expressive language is an important marker of delay in deficits in social communication, and is useful as an evaluative measure as it is easily observed and measured. Measuring the use of expressive language in naturalistic language contexts provides an indicator of the functional use of language for children with ASD. Functional use of language may discriminate children with ASD from those with language impairments (LI) only, because children with autism have difficulty with pragmatics of language (Tager-Flusberg et al., 2009). Understanding differences in spoken language between ASD and LI children could have important implications for differential diagnosis and for developing a better understanding of developmental trajectories.

Methods: Seventy children with LI participated (males=58, mean age= 30.1 months). Data were selected from a study examining the effects of Enhanced Milieu Teaching (EMT) for children with LI (Kaiser & Roberts, 2013). At entry, participants who received a positive screening for ASD were excluded. At follow-up, children were screened on the Autism Diagnostic Observation Schedule (ADOS). Seventeen children had scores indicating an ASD diagnosis. Regression analyses were completed to compare the expressive language of children with ASD to children with LI on naturalistic measures of spoken language to determine if children with an ASD diagnosis differed from children with an LI diagnosis only. Separate regression models were run, controlling for age, gender, ethnicity, and group assignment. Four measures of naturalistic language were used: total number of conversational turns and vocalizations collected using the Language Environment Analysis system (LENA) recording during one weekend day, the total number of words reported by the parent on the MacArthur-Bates Communicative Development Inventories (MCDI), the number of different words and total words during a 20-min naturalistic language sample, and the number of different words and total words during a 10-min parent-child interaction.

Results: The total number of words on the naturalistic language sample was a significant predictor of ASD (p=0.043), but the number of different words on the language sample did not reach significance (p=0.063). No other measures were statistically significant. There was a consistent pattern across four measures: number of different words in the language sample (p=0.063); total number of LENA child vocalizations (p=0.082), number of LENA conversational turns (p=0.074), and the number of words on the MCDI (p=0.093). The total number of words and the number of different words in the parent-child interaction were not significant predictors of ASD (p=0.177 and 0.40).

Conclusion: Measures of expressive word counts alone across three contexts did not differentiate children with ASD from children with LI. This indicates that functional-use measures of communication (e.g., initiations, commenting, responsiveness) may be necessary to evaluate the differences between these groups. However, the consistent pattern of measures approaching significance as predictors of ASD in these findings suggest that this area warrants further research. Developing efficient and effective strategies to better discriminate young children with LI from children with ASD is crucial in understating the communication patterns of these populations.

References:


Symposia Title: Social Communication Skills of Students With Disabilities including Autism Spectrum Disorders

Chair: Cathy Qi

Paper Title: Teaching Generalized Pretend Play and Related Behaviors to Young Children With Disabilities

Author(s): Erin Barton, Vanderbilt University

The United Nations High Commission For Human Rights asserted that play is the right of every child because it "is essential to the cognitive, physical, social, and emotional well-being of children and youth" (Ginsburg, the Committee on Communications, & the Committee on Psychosocial Aspects of Child and Family Health, 2007, p. 182). Play provides children with multiple opportunities to learn and engage with the environment, including promoting meaningful interactions across people and contexts (Barton, 2010; Lifter, Mason, & Barton, 2011; McConnell, 2002). Pretend play, in particular, affords multiple developmental benefits. Pretend play increases the likelihood of learning in inclusive preschool classrooms (Buysse, Wesley, Keyes, & Bailey, 1996) and provides contextually relevant opportunities to learn new skills across domains (Morrison, Sainato, Benchabban, & Endo, 2002). Children with disabilities play less often and demonstrate fewer varied pretend play behaviors than children with typical development. The two research questions guiding this study were: (a) Is there a functional relation between teachers' use of the instructional package (i.e., the system of least prompts and contingent imitation) and the unprompted, generalized, and novel pretend play behaviors of children with disabilities? (b) Is there a functional relation between teachers' use of the instructional package and the IEP (Individualized Education Program) goals?

Four children with disabilities were from an inclusive, non-profit preschool program within a university town in the Pacific Northwest were included in this study. An event recording system was used with child pretend play behaviors using the five-step process described in Barton and Wolery (2010). The first instructional condition focused on functional play with pretense behaviors (FPP); the second focused on symbolic play behaviors (SYM); and the third focused on IEP goals. The decision to intervene with subsequent behavior(s) was based on the presence of three consecutive sessions with more unprompted than prompted target behavior(s). A multiple probe design was used to examine the relation between teachers' use of the system of least prompts and contingent imitation and the acquisition, maintenance, and generalization of pretend play and related behaviors. The study met several quality indicators identified by Horner and colleagues (2005) for single subject research.

Results indicated the teachers' use of the intervention package was functionally related to increases in the children's frequency and diversity of pretend play and related behaviors. Children also maintained responses in sessions without prompts and generalized across toys and contexts. The findings replicate previous studies on adult prompting of pretend play and extend the literature by assessing generalization of children's pretend play across contexts and measuring intervention and implementation fidelity. This study supports previous research showing the positive effects of instructional packages using the system of least prompts to teach play (Lifter et al., 2005). The system of least prompts might be particularly effective for teaching play because it does not interrupt the play interaction and by starting with the natural prompt, the teacher can easily fade prompts. Also, the specific prompts used within the prompt hierarchy are selected based on the child's learning history and can be adapted based on the child's performance. Overall, this study provides a strong argument for engaging in systematic instruction of play, including pretend play, for children who do not display such behaviors.
The ability to initiate a conversation with a peer, to respond contingently to peers, and to take alternating turns has been found to relate to social acceptance in preschoolers (Diamond et al., 2008). Researchers have found that children who successfully orchestrate these skills with peers are preferred communication and play partners (e.g., Craig & Gallagher, 1986, Goldstein & Gallagher, 1992, Hadley & Rice, 1991). Interventions that focus on improving both social and communicative competence of children with disabilities are a key component of early intervention efforts (Stanton-Chapman & Snell, 2011). A number of interventions are available to ameliorate the social skills of children with disabilities. Many of these interventions explicitly teach social skills to this target population using intense direct instruction (Brown, Odom, & Conroy, 2001). For example, English, Shafer, Goldstein, and Kaczmarek (2005) taught children with developmental delays and typically developing peers to use social skills such as proximity to a peer, initiations, eye contact, and name use employing direct instruction strategies (e.g., modeling, practice, and discussion). For children with ASD and other developmental disabilities, this explicit instruction often is needed because naturalistic approaches are not sufficient to promote positive social interactions with peers (Brown, Odom, McConnell, & Rathel, 2008). The purpose of the current study was to evaluate the friendship skills (e.g., responding to a peer’s initiations; sharing toys; turn-taking) of children with ASD who participated in a social communication intervention using individualized social stories developed on an iPad along with modeling, practice, and discussion with an interventionist before peer play during center time.

A multiple baseline design across participants was used in this study. Three preschoolers with ASD (three males) participated in the study. Social stories were developed on an iPad and were based on each child’s language abilities and needed skills (e.g., responding to a peer’s initiations; sharing toys; turn-taking). After the stories were made, they were read once prior to the class’s center time where each child was observed. After the story was read with the children, the interventionist modeled, practiced, and discussed the friendship skills that were presented in the story with each child. Data was collected across baseline, intervention, maintenance and generalization phases. A total of 35 sessions of data collected for each participant.

Visual analysis and Percentage of None-Overlapping Data (PND; Scruggs, Mastropieri, & Casto, 1987) were utilized to evaluate the effectiveness of the intervention. The inter-observer reliability was 98% across 25% of all sessions. Social validity data was collected from teachers. Overall, they indicated that the individualized social stories were effective in promoting friendship skills. Procedural fidelity was 100% for all participants. The PND mean across participants and phases was .65 (SD = .21, Range = 0 - 2). The results showed that the individualized social stories were moderately effective to increase friendship skills across three participants during the intervention, maintenance, and generalization phases.

In conclusion, social stories developed on an iPad, along with modeling, practice, and discussion with an adult prior to peer play, are effective in increasing friendship skills across three students with ASD. The use of an iPad to teach the friendship skills appeared to motivate the children to engage with their peers during center time. These friendship skills were generalized to other settings as they continued to be used on the playground.
Increasing attention has focused on core features of the behavioral phenotype in fragile X syndrome with particular interest on the role of anxiety and attention and their interface with features of autism spectrum disorder across the developmental spectrum. In this series of talks, we will outline features of autism spectrum disorder in children with fragile X syndrome compared to a large sample of children with autism spectrum disorder (non-FXS) with a focus on items associated with attention. In addition, we will describe the early development of attention and its association with autism features and social anxiety. Finally, we will present data on the treatment of attention deficits using a novel approach. These papers represent multiple methods including physiological, genetic, and behavioral factors to examine dynamic system interactions over time in fragile X syndrome.
**Introduction:** It is well established that clinically significant levels of autism-related symptoms occur in a substantial proportion of children with Fragile X syndrome (FXS). While some studies have suggested that children with FXS and autism demonstrate highly similar phenotypes to children with "idiopathic" autism (Rogers et al., 2001), recent research indicates the need to examine specific ASD symptom dimensions in order to gain a more nuanced understanding of the similarities and differences in how ASD symptoms present across the two disorders (e.g., Wolff et al., 2012). Unfortunately, direct comparisons of FXS and ASD have been limited by relatively small samples of individuals with ASD. This has prevented adequate consideration of age, IQ, and language level, all of which are known to significantly affect the manifestation of autism symptoms in individuals with ASD. The current study combined data from two large existing datasets in order to be able to carefully compare domains that overlap in autism and FXS, as well as those that may show less convergence, when accounting for the influences of age, and cognitive and language ability across the two disorders.

**Methods:** Children were included if they had FXS (n=133) or ASD (n=1008) and a nonverbal IQ (NVIQ) score and Module 1, 2, or 3 of the Autism Diagnostic Observation Schedule (ADOS). We examined separately children who met ASD or autism cut-offs on the ADOS from those who scored in the non-ASD range. Preliminary analyses were conducted comparing ADOS calibrated severity scores (CSS) of boys with FXS who met ADOS cut-offs to boys with clinical diagnoses of ASD who met ADOS cut-offs. Subsequent analyses will examine individual ADOS item scores across the groups and will also include girls, as well as children with FXS and ASD who did not meet ADOS cut-offs. We will focus on items associated with attention.

**Results:** Initial comparisons showed large group differences between FXS and ASD on both NVIQ and age. We employed a propensity matching strategy to control for both of these variables simultaneously. Preliminary results indicated that boys with FXS who met ADOS cut-offs received significantly lower ADOS severity scores than children with ASD who met ADOS cut-offs, p<.01. Importantly, significant group differences were also found in distribution of ADOS modules across IQ groupings. For example, among children with FXS with NVIQs below 50, only 18% were administered a Module 1, whereas 86% of children with ASD in the same NVIQ category received a Module 1. This indicates potential differences in expressive language level between the groups not accounted for by NVIQ and highlights the need to consider Module in subsequent analyses.

**Discussion:** Results of this study will contribute information about the similarities and differences in autism symptoms between children with FXS and children with clinical diagnoses of ASD without FXS. Collaborative efforts between FXS and ASD researchers are essential in order to obtain samples large enough to account for all relevant matching variables. Carefully considering these variables allows for a more complete understanding of the overlapping aspects of the FXS and ASD behavioral phenotypes, which will inform research into similar and distinct mechanisms underlying the two disorders.

**Key References:**


**Symposia Title:** Attention, Anxiety, and Autism in Children With Fragile X Syndrome

**Chair:** Jane E. Roberts

**Paper Title:** Social Anxiety and Attention Modulation in Boys With Fragile X Syndrome

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Bridgette L. Tonnsen, University of South Carolina
Jane E. Roberts, University of South Carolina

**Introduction:** Eye-gaze avoidance during social interactions is well-documented in individuals with fragile X syndrome (FXS; Cohen, 1995; Hessel et al., 2006; Wolff et al., 1989). In addition, it appears that situations that are anxiety provoking, especially social situations, lead to hyperarousal that consequently lead to more gaze avoidance in this population (Belser & Sudhalter, 1995; Cohen et al., 1988). To our knowledge, no study as examined adaptive, or self-regulating, eye-gaze patterns in children with FXS in response to an unfamiliar person. During a stranger approach, a common response for typically developing (TD) children is to look to their parent for reassurance, which in turn helps them to regulate their emotional reaction (i.e., secure base behavior). Identifying the emergence of anxiety and the development factors associated with anxiety has implications for early identification and intervention. The purpose of this study was to examine eye-gaze in response to an unfamiliar person in children with FXS as compared to typically developing children from infancy into early childhood.

**Methods:** Participants were 57 boys with FXS and 59 boys with TD. Participants had a median of 3 observations that occurred between the ages of 9 and 59 months. The Stranger Approach episode from the Laboratory Temperament Assessment Battery (Goldsmith & Rothbart, 1996) was used to examine eye-gaze patterns in response to the approach of an unfamiliar person. The percent of time children spent looking to their mother, to the stranger, and away was coded from video by trained coders.

**Results:** Preliminary analysis indicate that boys with FXS and TD under 12 months had similar amounts of looking time to their mother, the stranger, and away. Between 12 and 23 months boys with FXS spent more time looking at their mother (F[1, 38] = 6.22, p = .02, eta2 = .14) or at the stranger (F[1, 38] = 4.18, p = .05, eta2 = .10), while boys with TD looked away more (F[1, 38] = 7.55, p = .01). Between 48 and 59 months, boys with TD looked toward their mother more (F[1, 35] = 9.39, p = .004, eta2 = .21) and boys with FXS looked away more (F[1, 35] = 10.41, p = .003, eta2 = .23). Growth modeling will be presented to characterize individual trajectories of change and summarize the mean rates of change within each group associated with these analyses.

**Discussion:** We found age-related trajectories of attention modulation during a social anxiety provoking situation in boys with FXS that differed from boys with TD. Unlike boys with TD—who look to their mother more over time—boys with FXS look away more in response to a stranger. While this behavior may reduce the fear associated with a stranger approach, it may not provide as much support in regulating their response as referencing their mother. Thus, the social and cognitive impairments associated with FXS may interact leading to less adaptive coping responses to stress.

**Key References:**


Introduction: Atypical visual attention has been well-documented among infants with older siblings diagnosed with autism ("A-SIBs") and is associated with increased risk for later autism diagnoses (see Zwaigenbaum et al., 2013, for review). Although atypical attention may contribute to the later socio-communicative deficits associated with autism, the neurobiological mechanisms of aberrant attention in A-SIBs are unclear. Heart-defined sustained attention (HD-SA) is a period of decelerative heart rate that has been used to index periods of attentional engagement in non-clinical infant samples (e.g. Casey & Richards, 1988). Despite documented patterns of atypical arousal in older children with autism (e.g. Bal et al., 2010), no studies have measured HD-SA as a potential biomarker of autism risk in A-SIBs. The present study integrated both behavioral looking and HD-SA to (1) contrast longitudinal profiles of attention across A-SIBs and low-risk controls (LRCs) between 6 and 12 months of age and (2) examine associations between attentional trajectories and clinical indicators of autism risk among ASIBs.

Methods: Data included 43 infants (21 A-SIBs, 22 LRCs; 4 females per group) assessed on 1-3 occasions (77 assessments; mean age=10.04 months, range=5.68-13.74 months). Behavioral looking and HD-SA were measured while infants passively viewed an engaging children's video. Dependent variables included proportion of time attending to the screen, overall interbeat interval (IBI) and respiratory sinus arrhythmia (RSA), and three features of HD-SA: proportion time in HD-SA, average IBI deceleration, and average IBI standard deviation (SD). The Autism Observational Scale for Infants (AOSI) Total Score measured clinical risk factors for autism at 12 months of age. Analyses were conducted using multilevel modeling, controlling for mental age (12 month Mullen Scales of Early Learning Standard Score).

Results: Both behavioral attention and HR-SA varied across groups. Although LRCs demonstrated decreased attentiveness over time, attentiveness increased with age in A-SIBs (B =-0.04, p=.003). Similarly, proportion of time in HD-SA decreased in the LRC group but remained relatively stable over time in A-SIBs (B =0.04, p=.03). The ASIB group also exhibited lower RSA across ages (B =-0.19, p=.04). Groups did not differ in overall IBI or remaining features of HD-SA. However, among A-SIBs, higher AOSI scores were associated with less robust longitudinal increases in overall IBI (B =-0.02, p=.05), as well as decreases in IBI deceleration (B =-0.17, p=.04) and SD (B =-0.18, p=.01) during HD-SA. Thus across analyses, A-SIBs displayed atypical longitudinal increases in behavioral and heart-defined attention, and higher autism risk at 12 months was predicted by increasingly shallower and less-variable heart rate during HD-SA over time.

Discussion: The first step to early identification and treatment of autism is fine-tuned characterization of the nature and course of atypical development. Our data suggest atypical age-related patterns of both behaviorally and physiologically-indexed attention in A-SIBs between 6 and 12 months of age. Notably, longitudinal patterns of heart rate during HD-SA predicted clinical autism symptoms at age 12 months, warranting further exploration of HD-SA as a potential biomarker of autism risk in infants.

Key References:


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**Symposia Title:** Attention, Anxiety, and Autism in Children With Fragile X Syndrome

**Chair:** Jane E. Roberts

**Paper Title:** Attention Training for Children With an Intellectual Disability: A Double Blind Randomized Controlled Trial

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**Introduction:** Attention is central to cognitive development and has been highlighted as a predictor of academic achievement and a facilitator of social inclusion. In the case of children who are vulnerable to learning impairments due to intellectual disability (ID), attention difficulties will likely exacerbate an already compromised cognitive system. Treatment options for improving attention skills in individuals with ID are sparse. Currently the most common intervention is psychostimulant medication and whilst the short term effects of drugs such as methylphenidate are well documented, less is known about the long term impacts of prolonged use. In light of the shortfalls of current interventions, recent investigations have suggested that cognitive training may provide an adjunct to pharmaceutical interventions and a riposte to the concept that cognitive impairments are permanent. This paper reports the first attempt to use cognitive training paradigms in children with ID.

**Methods:** In a double blind randomized controlled trial, 80 children with ID (Mage=8.02, range 4 to 10 years, IQ<75) were randomly assigned to an adaptive attention training program (intervention) or a non-adaptive control program. The intervention (Training Attention and Learning Initiative; TALI) consisted of a computerized program that ran on a tablet similar to an iPad. The program was developed specifically for children with ID, and incorporated selective attention, sustained attention and attentional control tasks. The intervention ran for 5 weeks and consisted of 25 sessions, lasting 20 minutes each. Children were assessed on a range of standardised and tailored before the intervention, immediately after the intervention and 3 months after the intervention. Both parent and teacher reports of inattentive behaviour were obtained.

**Results:** In this abstract we report preliminary results from ongoing data collection (n=33). Paired-samples t tests revealed that children in the intervention group showed significant improvements in performance on complex selective tasks immediately after training, (t (15) = -3.25, p<.01). Although improvements were not observed in other attentional processes immediately after training, improvements in basic selective attention (t (15) = -2.85, p<.05) and sustained attention (t (15) = -2.20, p<.05) were evident at the 3 month follow up. No improvements were observed in the control group on any attention task either immediately after training or at follow up. Behavioural measures of inattentive and hyperactive behaviour completed by parents and teachers indicated improvements in the intervention group after training, however these improvements did not reach significance.

**Discussion:** The targeted intervention produced improvements in core attentional processes in children with ID when compared to a control program. Importantly these results emphasise the potential of these training paradigms and offer an alternative to pharmaceutical interventions in individuals who are 'at risk' or already vulnerable to attention difficulties.

**Key References:**

