**Symposium Title:** Experiences, Health Outcomes, and Health Care Utilization of Children of Mothers with Intellectual and Developmental Disabilities

**Chair:** Monika Mitra

**Overview:** The four presentations included in this symposium address the experiences and health care outcomes, utilization and costs of infants and children of parents with disabilities, including those with intellectual and developmental disabilities. The presentations by Parish et al, Clements et al, and Mitra et al, use population-based data to examine the differences in health outcomes and utilization among newborns and infants of mothers with intellectual and developmental disabilities. Lightfoot and DeZelar address the disparities in outcomes and experiences of children in foster care who have parental disability as a reason for removal. Collectively, the four abstracts highlight the need for a greater understanding of the needs of parents with disabilities and their children, and for the development of interventions to address these disparities.

**Paper 1 of 4**

**Paper Title:** Population-Based Study of Hospital Utilization and Costs of Infants of Women with Intellectual and Developmental Disabilities

**Authors:** Monika Mitra\(^1\), Susan Parish\(^2\), Jianying Zhang\(^3\), Karen Clements\(^3\)\(^4\)

**Introduction:** There is a substantial body of literature on the health and well-being of parents of children with intellectual and developmental disabilities. In recent years a body of work on the maternal and birth outcomes of women with disabilities including women with intellectual and developmental disabilities has emerged. Using national population-based data Parish et al. (2015) found that women with intellectual and developmental disabilities in the United States were more likely to experience pregnancy complications and more likely to experience poor maternal outcomes. Similarly Mitra, Parish, Clements, Cui, & Diop (2015) using population-based data from a longitudinal linked database found similar results. They found that deliveries to women with intellectual and developmental disabilities were to those who were younger, less educated, more likely to be black and Hispanic, and less likely to be married. They were less likely to identify the father on the infant's birth certificate, more likely to smoke during pregnancy, and less likely to receive prenatal care during the first trimester compared to deliveries to other women. This study extends the earlier studies by examining the outcomes and health care utilization and cost of infants up to one year of age born to mothers with intellectual and developmental disabilities.

**Methods:** Data from the 1998-2010 Massachusetts Pregnancy to Early Life Longitudinal database were analyzed to identify infants born to Massachusetts women with intellectual and developmental disabilities. Bivariate analyses were conducted to compare health care utilization and costs between infants born to women with intellectual and developmental disabilities and women in the general obstetric population. Chi-square statistics were used to compare the distribution of characteristics of the infants to the two groups of women. Wilcoxon tests will be used to compare median hospital utilization costs between infants of women with intellectual and developmental disabilities and other women.

**Results:** Of the 950,035 infants born in Massachusetts between 1998 and 2009, 1,160 (0.1%) infants were to women with intellectual and developmental disabilities. Infants born to women with intellectual and developmental disabilities were more likely to be born of low birth weight (13.5% compared to 7.9% of other women, p<0.001). Infants born to mothers with

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1. Brandeis University
2. Lurie Institute for Disability Policy
3. University of Massachusetts Medical School
4. Center for Health Policy and Research
intellectual and developmental disabilities were also more likely to have emergency department visits (31.6% vs 21.2%), observational stays (6.6% vs 3.1%), and non-delivery hospitalizations (15.3% vs 9.1%) compared to infants born to other mothers (p<0.001) in the first year of their life. There were no significant differences in the average length of observational and hospital stays between the two groups of infants. However there were significant differences in the median cost of emergency room, observation stays and hospital stays between infants of mothers with intellectual and developmental disabilities compared to those of other mothers.

Discussion: These findings of this study highlight significant disparities in the health care utilization of infants of mothers with intellectual and developmental disabilities compared to mothers without intellectual and developmental disabilities. The findings of the study point to a need for further research is needed to understand the higher rates of hospital visits among these infants and the development of appropriate interventions and guidelines to address these disparities.

References/Citations:


Paper 2 of 4

**Paper Title:** Medical Conditions among Massachusetts Infants Born to Women with Intellectual and Developmental Disabilities

**Authors:** Karen Clements\(^3\) \(^4\), Monika Mitra\(^1\) \(^2\), Jianying Zhang\(^3\), Susan Parish\(^1\) \(^2\)

**Introduction:** A growing body of research demonstrates that children born to women with intellectual and developmental disabilities (IDD) are at risk for adverse birth outcomes, including low birthweight, preterm birth and perinatal death (Parrish et al, 2015, Mitra et al, 2015). More limited data suggests that these infants are at risk for poor health and developmental delays (Hindmarsh G et al, 2015, Emerson et al, 2014). Data regarding medical diagnoses among infants born to women with IDD, however, is lacking. The objective of this analysis is to compare the prevalence of medical diagnoses at birth hospital discharge among infants born to women with and without IDDs.

**Methods:** Data from the 1998-2009 Massachusetts Pregnancy to Early Life Longitudinal database were analyzed to identify infants born to Massachusetts women with IDDs. Clinical Classification Software (CCS) grouped ICD-9-CM codes from hospital discharge data into diagnostic categories. Chi-square statistics compared the prevalence of diagnoses, by CCS category, between infants born to women with and without IDDs.

**Results:** During the study period, 1,160 (0.1%) of the 950,035 infants born in Massachusetts were born to women with IDDs. At birth hospital discharge, infants born to women with IDD had a higher prevalence of diagnoses in all CCS categories compared with infants born to mothers without IDD. The most common diagnostic categories were congenital anomalies (0.19 diagnoses/infant, moms with IDD vs 0.09, moms without IDD, p < 0.0001), diagnoses of the nervous system and sense organs (0.04 vs. 0.02, p = 0.009) and mental illness (0.03 vs. 0.02, p < 0.0001). The most frequent diagnoses among infants born to mothers with IDD were congenital pigmented anomalies of the skin, patent ductus arteriosus, and drug withdrawal syndrome. These diagnoses were also the most frequent among infants born to mothers with no IDD.

**Discussion:** These findings suggest that infants of mothers with IDD may have an elevated prevalence of medical conditions diagnosed shortly after birth. There does not appear to be any differences in the distribution of diagnoses between infants born
to mothers with and without IDD. Future research will evaluate long term costs associated with these diagnoses and identify additional medical conditions diagnosed during the first three years of life.

References/Citations:


Paper 3 of 4

Paper Title: Fetal Outcomes among U.S. Women with Intellectual and Developmental Disabilities

Authors: Susan Parish1 2, Monika Mitra1 2, Ilhom Akobirshoev1 2, Leah Igdalsky1 2

Introduction: To date, there is little existing research on fetal outcomes with a population-based sample of U.S. women with intellectual and developmental disabilities. However, research from other countries that children born to mothers with intellectual disabilities face increased risk of adverse fetal outcomes. A 2012 Swedish study found that children born to mothers with intellectual and developmental disabilities were more often stillborn or died perinatally than children born to mothers without intellectual and developmental disabilities (Höglund, Lindgren, & Larsson, 2012). An Australian cohort study found that 28% of children in their sample born to mothers with intellectual disabilities were born premature, and 22% had low birth weights (McConnell, Mayes, & Llewellyn, 2008).

This study sought to determine the nationwide number of deliveries occurring in women with developmental disabilities and compared the rates of deliveries complicated by adverse fetal outcomes, including preterm birth, fetal growth restriction, and stillbirth in women with intellectual and developmental disabilities to the general obstetric population.

Methods: Data were drawn from the Nationwide Inpatient Sample (NIS) of the Health Care and Cost Utilization Project which is nationally representative of hospital discharges. Data from 2008-2011 was used to increase the sample size and power of the analysis. Women with intellectual and developmental disabilities were identified by ICD-9 diagnostic codes. We compared preterm birth, fetal growth restriction or low birth weight, and stillbirth for women with and without IDD across age, race, insurance payer, region of hospital, location and teaching status of the hospital, ownership of the hospital, and median household income for the patient’s zip code. The sample consisted of 6,500 hospitalizations of women with intellectual and developmental disabilities and 15,985,183 hospitalizations of women without intellectual and developmental disabilities, weighted.

Results: Women with IDD had worse outcomes for all fetal outcomes than the general obstetric population. Namely, women with IDD had about two -fold higher rates of preterm birth, low birth weight, and stillbirth than the general obstetric population. Women with IDD compared to the general obstetric population were significantly more likely to have worse fetal outcomes, including having preterm birth (OR=1.74; 95%CI: 1.47-2.06, p<0.001), low birth weight (OR=1.93, 95%CI: 1.44 - 2.59, p<0.001), and stillbirth (OR=2.18, 95% CI: 1.35 - 3.53).
Discussion: Children born to mothers with intellectual disabilities are at increased risk of negative fetal outcomes. Targeted interventions are needed to address these deleterious outcomes.

References/Citations:

Paper 4 of 4

Paper Title: How Children of Parents with Disabilities Fare in the Child Welfare System

Authors: Elizabeth Lightfoot\(^5\), Sharyn DeZelar\(^5\)

Introduction: There has been a growing concern about the involvement of parents with disabilities in the child welfare system, though there is little research on how child welfare agencies are serving parents with disabilities and/or their families. While nationally available data on parental disability in child welfare is limited, the Adoption and Foster Care Reporting System (AFCARS), the federal reporting system that collects case-level data on all children in foster care, does include a removal reason regarding a caretaker's "physical or emotional illness or disabling condition". The purpose of this study is to examine how states use the parental disability removal reason and the experiences and outcomes of children in foster care who have parental disability as a removal reason.

Methods: This study used 2012 data from AFCARS, in which 19.5% of foster children had at least one removal reason of parental disability, and 5.6% had parental disability as their sole removal reason. Descriptive statistics were used to explore the variations in states' use of parental disability as a removal reason, both by itself and in conjunction with other removal reasons. Logistic regression was used to explore how the parental disability removal reason correlated with type of placement, case plan of reunification, and reunification upon discharge. T-tests were used to compare children with and without the parental disability removal reason in regards to length of stay in current placement, and total days in foster care.

Results: There is wide variation in the states' use of the parental disability removal reason. Six states used parental disability as at least one removal reason in more than 30% of their cases, and five states had over 10% of their cases where parental disability was the sole removal reason. Children who had parental disability as a sole removal reason were 1.34 times more likely to be in non-relative foster care than were those without parental disability as a sole removal reason. Children who had parental disability as one of several removal reasons spent an average of 100 days longer in their current foster care setting and 116 more total days in foster care than those without parental disability as a removal reason. Children with parental disability as a sole removal reason spent an average of 205 days more in their current setting and 240 more total days in foster care than those without disability as a sole removal reason. Those with parental disability as one of several removal reasons were 33% less likely to have a case goal of reunification and 22% less likely to be reunified upon case closure, and those with parental disability as a sole removal reason were 32% less likely to have a case plan of reunification and 50% likely to have reunification as an outcome upon case closure.

Discussion: In this study, foster children who had parental disability as a removal reason had different foster care experiences and different child welfare outcomes than foster children without parental disability as a removal reason. While the AFCARS

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removal reason of parental disability is not a proxy for parental disability and is unevenly used by child welfare agencies, the study points to a need for closer attention to parental disability within the child welfare system to ensure appropriate services for parents with disabilities and their children.

References/Citations:

Symposium Title: The Delivery of Evidence-Based Social Skills for Youth and Young Adults with Autism Spectrum Disorders: Theory, Research and Practice

Chair: Elizabeth Laugeson¹

Discussant: Jan Blacher¹ ²

Overview: Participants attending this symposium will learn about current evidence-based practices in social skills intervention. Theoretical considerations, translational as well as outcome research conducted across settings will be discussed and variables implicated in treatment outcome will be analyzed.

Paper 1 of 4

Paper Title: Selecting Informants to Assess Social Functioning and Treatment Outcome for Adolescents with Social Challenges

Authors: Elina Veytsman¹, Crystal Ferrendelli¹, Shannon Bates¹, Elizabeth Laugeson¹

Introduction: Assessment of social functioning of youth with social challenges is complicated by conflicting informant perceptions. For youth with ASD, self-report of symptoms of psychiatric diagnoses should be interpreted with caution (Mazefsky et al. 2011), as this population has shown poor diagnostic agreement with parents (Storch et al. 2012), underscoring the need for multiple informants, including teachers and therapists. Research shows concordance rates between parent and adolescent report are widely heterogeneous, dependent upon the instrument, the disorders under investigation, and the informant characteristics (Mazefsky et al. 2011). Understanding the discrepancy between parent, teacher, and self-report of social functioning and treatment outcome among youth with social challenges is critical for determining the most reliable informants. The current study examines this discrepancy following a 14-week evidence-based social skills intervention for adolescents with social difficulties in order to determine if perceptual differences change over time.

Methods: Participants included 343 adolescents with social challenges referred for social skills training in outpatient and school settings. Among the clinic sample, participants included 237 adolescents (males=178; females=59) 11-18 years of age (M=13.8, SD=2.25) with diagnoses of ASD (n=126), ADHD (n=52), Anxiety (n=8), and other (n=51) who attended 14 sessions of a weekly 90-minute social skills group with their parents using the Program for the Education and Enrichment of Relational Skills (PEERS®; Laugeson & Frankel 2010). Within the school sample, participants included 106 adolescents (males=86; females=20) 11-18 years of age (M=15.08, SD=1.82) with ASD who received daily teacher-facilitated social skills instruction in the classroom using the PEERS® school-based curriculum (Laugeson 2014). In order to assess perceptual differences of social functioning, adolescents and parents completed the Social Anxiety Scale (SAS; La Greca 1999), Quality of Socialization Questionnaire (QSQ; Frankel & Mintz 2008), and Empathy Quotient (EQ; Baron-Cohen 2004) at pre and post-test. Parents and teachers also completed the Social Skills Improvement System (SSIS; Gresham & Elliott 2008) and Social Responsiveness Scale (SRS; Constantino 2005) pre and post-treatment. Paired sample T-tests and Pearson product-moment correlations were conducted to examine informant perceptions of adolescent social functioning across settings, and Bonferroni adjustments were made.

Results: Results reveal moderate and significant correlations between parent, adolescent and teacher report for measures of social functioning. However, there were significant differences (p <.001) between parent and adolescent report of social anxiety and engagement, and parent and teacher report of social skills and autism symptoms at baseline and post-treatment. These differences decrease at post-treatment across measures in both samples, signifying increased agreement between informants.

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following intervention. Adversely, differences in adolescent and parent report of social engagement measured by the QSQ significantly (p < .001) increase at post-treatment in the school-based sample.

**Discussion:** This study highlights the complexity of using multiple informants in the assessment of social skills across settings. The gap between parent, teacher, and adolescent perceptions of adolescent social functioning lessens over time and treatment in both samples. However, the discrepancy between adolescent and parent report of social engagement increases post-treatment in the school sample, which may be explained by less involvement of parents in school-based interventions. The results highlight the need for multiple informants in social skills assessments.

**References/Citations:**


**Paper 2 of 4**

**Paper Title:** Examining Delivery Models for Evidence-Based Social Skills Intervention for Adolescents with Autism

**Authors:** Courtney Bolton1 3, Elizabeth Laugeson1 4

**Introduction:** Research indicates that an increasing number of youth lack the appropriate interpersonal skills required for developing peer connections, making evidence-based social skills intervention paramount to their success (Knoff, 2002). An analysis of social skills programs for individuals with social challenges aligned with the core deficits of autism spectrum disorders (ASD) identified schools as the most common research setting for intervention highlighting the need to examine outcomes in other settings, such as outpatient and community locations (Reichow & Volkmar, 2010). Further research is needed on the mechanisms of treatment outcome across settings. The current study examines the pretreatment variables associated with positive gains in social skills in both outpatient and school settings. This presentation investigates findings following an evidence-based social skill intervention facilitated in two independent settings to compare findings from a psychologist-facilitated, parent-assisted group implemented in an outpatient setting and a teacher-facilitated group conducted in an applied school setting.

**Methods:** The study examined 106 adolescents with ASD ages 11-18 (M = 15.08, SD = 1.82) who received social skills instruction through their nonpublic school and 237 outpatient adolescents with ASD ranging in age from 11-18 years (M = 13.8, SD = 2.25) who attended weekly outpatient social skills training sessions along with their parents. Treatment in both settings included a 14-week manualized intervention, UCLA Program for the Education and Enrichment of Relational Skills (PEERS®), a social skills curriculum for youth with ASD. To examine treatment outcome, adolescent participants completed pre and post-test measures, including the Piers-Harris Self-Concept Scale - Second Edition (PHS2; Piers, Harris, Herzberg, 2002), while parents completed the Social Skills Improvement System (SSIS; Gresham & Elliot, 2008) and the Social Responsiveness Scale (SRS; Constantino & Gruber, 2005). Data was analyzed using SPSS 22 to conduct paired t-test to examine pre and post differences. In addition, multiple regression analysis was conducted to investigate pretreatment variables associated with increases in social skills outcomes in the two distinct settings.

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4 The Help Group-UCLA Autism Research Alliance
**Results:** Participants in both settings achieved significant, positive gains in social skills on the PHS-2 ($p \leq .000$), SSIS ($p \leq .000$), and SRS ($p \leq .000$). Data analysis reveals distinct pretreatment variables that are able to account for the variance in social skills gains following the intervention in outpatient and school settings. Social responsibility, popularity and control were found to significantly ($p \leq .000$) explain the variance in outcome scores in the outpatient setting. The three-factor model accounted for 27% of the variance with outpatient participants. Different pretreatment variables, including social communication, perceived intellect and school status and friendship quality significantly ($p \leq .000$) accounted for 44% the variance in outcomes for the school-based setting.

**Discussion:** Data reveals that participants in both settings achieved positive social skills gains, but distinct pre-treatment variables acted as significant predictors for social skills in the different settings. Although consistent with previous research conducted in a clinic setting, this study highlights different pretreatment variables that should be further investigated in distinct settings. Future research should be conducted on varying mechanisms for change and moderators for treatment success based on setting.

**References/Citations:**

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**Paper 3 of 4**

**Paper Title:** Investigating the Relation between Friendship Quality and Social Engagement Following a School-Based Social Skills Intervention

**Authors:** James Yang\(^1\)\(^4\), Lara Tucci\(^4\)\(^5\), Yasamine Bolourian\(^2\)\(^4\), Elizabeth Laugeson\(^1\)\(^4\)

**Introduction:** Impaired social functioning among youth with Autism Spectrum Disorder (ASD) may lead to lack of social engagement and poor friendship quality (Ormond, Krauss, & Seltzer, 2004; Bauminger & Kasari, 2000). Greater friendship quality has been associated with fewer social problems, and less internalizing and externalizing problem behaviors. Having at least one close friend has been shown to be a protective factor against peer victimization and better overall adjustment in adolescents (Waldrip, Malcolm, & Jensen-Campbell, 2008). Children with ASD often report having friendships in school settings, but these friendships are often focused on circumscribed interests with little outside social engagement (Church, Alisanski, & Amanullah, 2000). Despite the growing research examining the social functioning of school-aged children with ASD, little is known about the friendship quality and social engagement in older youth. The present study seeks to address this research gap by examining the relationship between friendship quality and social engagement among adolescents with ASD in school settings.

**Methods:** Participants included 106 adolescents (males=86; females=20) ranging from 11-18 years of age ($M=15.08$; $SD=1.82$) at The Help Group’s Village Glen School, a nonpublic school for youth with ASD. Participants were involved in a larger treatment outcome study investigating the effectiveness of the PEERS® school-based curriculum (Laugeson 2014). In order to examine the

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\(^5\) Pepperdine University
relationship between friendship quality and social engagement, adolescents completed the Friendship Qualities Scale (FQS; Bukowski, Hoza, & Bolvin, 1994) at baseline. The FQS measures constructs of friendship including companionship, conflict, and security. To assess social engagement, adolescents completed the Quality of Socialization Questionnaire (QSQ; Frankel & Mintz, 2008) prior to treatment. The QSQ measures the number of hosted and invited get-togethers and degree of conflict during get-togethers in the previous month. Pearson correlation coefficients were calculated to examine the relationship between friendship quality on the FQS and social engagement using the QSQ.

**Results:** Results demonstrate moderate positive correlation between the FQS baseline scores and baseline scores on the QSQ frequency of invited get-togethers (p<.05), and a moderate inverse relationship with baseline scores on the FQS and the QSQ conflict scale (p<.05). In addition to these findings, the companionship subscale on the FQS is correlated with higher baseline scores on the QSQ frequency of invited get-togethers (p<.001), and the QSQ frequency of hosted get-togethers (p<.001). Higher baseline companionship scores on the FQS are also correlated with lower baseline scores on the QSQ conflict scale (p<.002).

**Discussion:** These findings suggest that adolescents with ASD that exhibit higher friendship quality and demonstrate higher companionship are more likely to be socially engaged with their peers and experience less conflict within their friendships. These results suggest that when the treatment priority is to increase social engagement and decrease conflict, the need for more targeted interventions to improve friendship quality and companionship may be warranted. 

**References/Citations:**

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**Paper Title:** Examining PEERS for Young Adults: Improvements in Social Motivation and Relations to Neural Activity

**Authors:** Bridget Dolan, Alexander Barrington, Dylan Snyder, Alana McVey, Amy Van Hecke

**Introduction:** Social skills deficits among adults with ASD lead to isolation and a lack of friendships. The Program for the Education and Enrichment of Relational Skills for Young Adults (Gantman et al., 2012) is an empirically based, caregiver-assisted treatment that teaches young adults with ASD how to make and keep friends. There are no published studies that have evaluated PEERS for Young Adults and its effects on brain function. Thus, this study seeks evaluate intervention influences on brain function, as assessed via EEG, and examine if neural changes relate to improvements in social functioning and mental health.

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6 Marquette University

Page 4 of 5
**Methods:** Analyses included 25 young adults (18-26 years old) with ASD. All participants had a verbal IQ > 70 and diagnoses were confirmed with the ADOS. The intervention was the 16-session PEERS for Young Adults. Measures were taken at pre- and post-intervention and included: 1) the Social Responsiveness Scale (caregiver report; SRS: Constantino, 2005); 2) the Beck Depression Inventory (BDI: Beck & Steer, 1987); and 3) a 3-minute continuous, resting state EEG recording.

**Results:** For the experimental group, caregivers at post-treatment reported significant improvements in social motivation on the SRS (F(1, 23) = 6.10, p < .05). There was a significant group by time by location interaction within the beta frequency band (F(1, 23) = 4.16, p < .05), with the experimental group demonstrating a significant decrease in left temporal-parietal beta power at post-treatment. This decrease in beta band activity was related to improvements in social motivation following PEERS (r = .49, p < .01). Group differences did not emerge on the BDI.

**Discussion:** Young adults receiving PEERS demonstrated a significant improvement in caregiver-reported social motivation. This finding was related to a decrease in left temporal-parietal beta power, which research suggests is indicative of better neural control (Orekhova et al., 2007). Greater neural control in this region of the brain, which processes social input, may explain the relationship with increased motivation to engage socially with peers. The results from this study add to the minimal literature that has examined efficacious social skills interventions for adults.

**References/Citations:**
Symposium Title: Health Behaviors and Outcomes in Females with an FMR1 Mutation

Chairs: Anne Wheeler\textsuperscript{1,2}, Melissa Raspa\textsuperscript{1}

Overview: Women with an FMR1 mutation are at high risk of having offspring with fragile X syndrome (FXS), the leading hereditary cause of intellectual disability and most common single gene cause of autism. In addition females with a pre or full mutation face additional health (e.g. fragile x-associated primary ovarian insufficiency), cognitive (e.g. impaired cognitive functioning in women with a full mutation; possible executive dysfunctions in women with a premutation) and psychosocial (e.g. increased chance of depression, anxiety, autism symptoms) risks which may comprise health and reproductive decision making. This symposium will feature 3 papers that focus on health behaviors and health related outcomes for females with an FMR1 pre or full mutation, with a discussion focused on how these behaviors and outcomes may impact reproductive decision making for these women.

Paper 1 of 3

Paper Title: Decision-Making and Health Behaviors in Girls and Women with Fragile X Syndrome

Authors: Kylee Miller\textsuperscript{2}, Anne Wheeler\textsuperscript{1,2}, Melissa Raspa\textsuperscript{1}

Introduction: There has been an increasing emphasis on the health outcomes of individuals with intellectual and developmental disabilities (IDD), especially the effects of weight on emotional, cardiovascular, and cognitive functioning (Melville et al, 2007). Females and individuals with mild ID may be at particular risk for obesity and other health related issues (Stancliffe et. al, 2011). Opportunities and ability to make minor and major life decisions is based in part on an individual’s ability to understand the decision as well as on previous opportunities and experiences. This presentation will provide preliminary results from a study exploring the relationship between decisions regarding healthy lifestyle and food choices, cognitive functioning, and weight in a sample of adolescent and adult females with fragile X syndrome (FXS).

Methods: Participants completed several direct assessments including the Sanford-Binet-V to assess broad IQ and the Delis-Kaplan Executive Function System to measure cognitive flexibility, inhibition, and abstract thinking; as well as a nutrition decision questionnaire adapted from the Healthy Eating Adds Up curriculum (Johnson, 2012). Height and weight were used to calculate body mass index (BMI). In addition, guardians completed questionnaires about adaptive behaviors using the Scales of Independent Behavior-Revised, anxiety using the Anxiety, Depression, and Mood Scale, and an assessment of the participants’ decision-making experiences. The decision-making questionnaire was a caregiver rating of the participant’s experience making daily decisions as well as bigger life decisions. Particular attention was paid to items related to health and nutrition decision making.

Results: This presentation will provide data on over 50 females with FXS. Preliminary analysis on 39 females with full mutation FXS suggest the majority are able to make basic decisions independently or with minimal help [e.g. what to cook=95%; select snack items=92%; when to exercise=95%; what to order from a menu=69%]. Multiple regression was conducted to determine the best linear combination of age, IQ, BMI, anxiety, cognitive flexibility, inhibition, abstract thinking, and decision making experiences for cooking, choosing snacks, exercising, ordering from a menu for predicting nutrition decision making. This combination of variables did not significantly predict healthy nutritional decision making F (13,14) =1.420, p=.261. However, cognitive ability and inhibition were significant predictors of healthy nutritional decisions.

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\textsuperscript{2} University of North Carolina at Chapel Hill
Discussion: While the preliminary analysis did not provide a significant model to predict healthy nutritional decision making in females with FXS, we did find significant relationships with IQ and inhibition. Not surprisingly, preliminary analyses indicate that a higher IQ is associated with better nutritional decisions, while disinhibition, or a lack of restraint, is associated with fewer good nutritional decisions. The presentation will provide updated analysis with the full sample and discuss nutrition and other health related decision making in females with FXS and implications for long term health in these women.

References/Citations:


Paper 2 of 3

Paper Title: Prenatal, Birth, and Postnatal Experiences of Women with an FMR1 Mutation

Authors: Anne Wheeler¹², Melissa Raspa¹, Amanda Wiley¹

Introduction: Accumulating research suggests an increased risk for greater medical and emotional co-morbidity and physical health symptoms among women with an FMR1 expansion (Wheeler, Bailey, et al., 2014). However, although women with the premutation are known to be at risk for developing Fragile X Primary Ovarian Insufficiency (FXPOI), little is known about their actual experiences with reproduction, pregnancy, birth and postpartum experiences. Results from a large national US survey are described with regard to reproductive decision making, pregnancy, birth and labor, and postpartum experiences, including breastfeeding.

Methods: This presentation summarizes findings from a large internet and phone based survey of 829 families affected by fragile X. 640 adult women with an FMR1 mutation reported on their experiences with various reproductive experiences including challenges with reproduction (e.g. use of reproductive assistance), pregnancy (e.g. preeclampsia), birth and delivery (e.g. length of labor), and the postpartum period (e.g. postpartum depression and breastfeeding). Questions about birth/labor and breastfeeding were asked for each child in the family, allowing a comparison between experiences with birth for children with a full mutation, premutation, and non-affected.

Results: As previously reported (Wheeler, Raspa et al., 2014), respondents with the premutation were twice as likely to have used any reproductive assistance to conceive (prior to knowing their FX status) than respondents with the full mutation or the national average. They were also more likely to experience preeclampsia than would be expected in the general population. New analyses revealed that most pregnancy lengths were at-term, less than 2% were pre-term and 4% were over 40 weeks. Most women breastfed for at least 3 months, although a quarter of women did not breastfeed at all. Of the women who did not breastfeed or stopped before 6 weeks, 24% did so due to difficulties experienced by the women (e.g. insufficient milk production, too painful); while 30% reported the child’s difficulties (e.g. poor suck reflex, reflux) were the primary reason for stopping breastfeeding. Children with the FM were more likely than children with a PM or those who were non-affected to have difficulties breastfeeding due to having a poor reflex; children with a PM were more likely to have needed it to be very quiet in order to breastfeed than children with a FM or those who were not affected. Children described as having more sensory issues later in life were also more likely to have bitten, needed it to be dark, pulled away from parental touch, and spit up more than
usual during breastfeeding. Children with more attention, hyperactivity, anxiety, autism, and thinking, learning, and reasoning problems were more likely to have mothers who did not breastfed or who reported more difficulty breastfeeding.

**Discussion:** Women with an FMR1 premutation generally have typical experiences with pregnancy, birth and labor, and the postpartum period. However, children with a FM may be less likely to be breastfed due to challenges with poor suck reflex and very early sensory concerns. These are issues which need to be further studied in order to best support women and young children with FMR1 mutations.

**References/Citations:**

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**Paper 3 of 3**

**Paper Title:** Reproductive Health Behaviors of Females with Fragile X Syndrome

**Authors:** Melissa Raspa¹, Anne Wheeler¹, Amanda Wiley¹

**Introduction:** Females with fragile X syndrome, the most common inherited form of intellectual disability, are at risk for passing on the genetic condition to their offspring. However, most studies to date have focused on the reproductive risk and health of women with the premutation, especially those with fragile X-associated primary ovarian insufficiency (FXPOI). Few studies have examined the reproductive health behaviors and knowledge among females with the full mutation.

**Methods:** As part of a large national survey, families who have a child with FXS answered questions about reproductive health behaviors of their daughter with the full mutation for which they were legal guardians. Respondents chose to complete a web-based survey or a phone interview and were directed to modules based on age and genetic status. Ninety two percent of families were Caucasian, 3% were African American, 3% were Hispanic, and 3% were from another racial or ethnic background. About two-thirds (66%) of the families had incomes of $75,000 or more. The majority of respondents were married (85%), employed (55%), and had a 4 year college degree or more education (62%). Families reported on 131 females with the full mutation (under 18 years = 89, 18 to 25 years = 19, over 25 years = 23).

**Results:** Less than half of females 15 years or older (45%) had been to an obstetrician-gynecologist (OB-GYN). Of those who had, most had their first visit between 18 and 20 years of age (38%). Females experienced a range of levels of comfort in visiting an OB-GYN, with 29% indicating their daughter was not at all comfortable, 24% a little comfortable, 33% somewhat comfortable, and 14% very comfortable. Females had been diagnosed or treated for a variety of reproductive health problems, including absent or irregular periods (15%), or other problems (19%). No females had been diagnosed or treated for infertility issues or early menopause. When asked about their knowledge of reproductive health, 50% fully understood anticipated changes in the body (e.g., menstruation, hair growth), 31% understood how pregnancy occurs, 28% understood birth control options, 68% understood the different between private and public sexually-related behaviors, and 41% understood the heritability of FXS. Regression models found that age (p < .0001) and family income (p = .0078) were predictive of a total knowledge score, but total co-occurring conditions, overall ability, and ability to interact were not statistically significant.
**Discussion:** The majority of studies to date have focused on reproductive health in women with the FMR1 premutation (Raspberry & Skinner, 2011; Sherman, 2000; Whittenberger et al., 2007). These data provide new information about the reproductive health behaviors and knowledge for females with the full mutation. In general, reproductive health behaviors and knowledge are associated with age and not functioning level.

**References/Citations:**
**Symposium Title:** Transactions during the Prelinguistic Period in Children Diagnosed with, or at High Risk for, Autism Spectrum Disorder

**Chair:** Tiffany Woynaroski

**Discussant:** Jana Iverson

**Overview:** Transactional theory suggests that parent and child factors interact in a dynamic way to support children's language learning. One instantiation of transactional theory proposes that, during the prelinguistic period, child vocalizations elicit parent linguistic responses that scaffold language development. This panel presents recent findings in support of this transactional model of spoken language learning in children who are diagnosed with, or at heightened risk for ASD, from research carried out at several different institutions, including Vanderbilt University, University of North Carolina at Chapel Hill, and University of Pittsburgh. Implications of the findings for research, theory, and clinical practice will be discussed.

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**Paper 1 of 4**

**Paper Title:** Empirical Support for a Transactional Model of Spoken Language Learning in Preschoolers with Autism Spectrum Disorder

**Authors:** Tiffany Woynaroski, Paul Yoder, Linda Watson

**Introduction:** There is a pressing need to explain individual differences in "useful speech" of young children with autism spectrum disorder (ASD) because learning to use spoken language to communicate early in life has been repeatedly linked with long-term outcomes in this population. Transactional theory suggests that models considering both parent and child factors, and the dynamic way in which such factors interact, will best account for individual differences in spoken language acquisition. One instantiation of transactional theory specifically suggests that early child vocalizations that are more "speech-like" or "complex" elicit parent linguistic responses, which in turn scaffold spoken language learning. We have previously confirmed this model in a group of young children with developmental disabilities not due to autism. The present study tests whether this model additionally applies to preschoolers with ASD.

**Methods:** We drew on extant data from a recent longitudinal investigation of useful speech development involving 87 preschoolers with ASD. An index of the complexity of children's vocalizations (i.e., consonant inventory in communicative vocalizations) was measured at Time 1 from the Communication and Symbolic Behavior Scales-Developmental Profile (CSBS-DP). Children's spoken language outcomes were measured 8 months later at Time 3 using an aggregate of scores from the CSBS-DP word scale, the Macarthur-Bates Communicative Development Inventories: Words and Gestures expressive vocabulary checklist and the number of different words produced in a semi-structured communication sample with an examiner. Midpoint parental linguistic mapping was measured at Time 2 (4 months after Time 1) in a 10 minute parent-child interaction. Mediation analysis with stochastic regression handling of any missing data points was used to test the statistical significance of the indirect effect of early child vocal complexity on later spoken language outcomes through mid-point parental linguistic mapping (Hayes, 2009).

**Results:** As expected, children's early vocal complexity predicted their later spoken language outcomes ($r = .61; p < .001$). Additionally, early child vocal complexity was positively associated with mid-point parent linguistic mapping ($r = .33; p < .001$), and mid-point parent linguistic mapping predicted later spoken language outcomes, controlling for early child vocal complexity ($r$

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1. Vanderbilt University
2. University of Pittsburgh
3. University of North Carolina at Chapel Hill
The indirect effect of early child vocal complexity on later spoken language outcomes through mid-point parent linguistic mapping was statistically significant (p = .03). Thus, mid-point parent linguistic mapping mediated the relation between early vocal complexity and later spoken language in our sample.

Discussion: These findings lend empirical support to the transactional theory of spoken language development in preschoolers with ASD. We confirmed that one child factor (early vocal complexity) and one parent factor (linguistic mapping) contribute in a dynamic manner to impact spoken language outcomes in children with ASD. This result suggests that we might best support spoken language acquisition in this population by targeting both the complexity of children's vocalizations and parents' responses to such prelinguistic child productions. Implications for theory, research, and practice will be discussed.

References/Citations:

Paper Title: Predictors of Growth in Diversity of Consonants Used in Communication Acts in Initially Nonverbal Children with Autism Spectrum Disorder

Authors: Paul Yoder¹, Linda Watson³, Tiffany Woynaroski¹

Introduction: Diversity of 10 key consonants (and their cognates) used in communication acts (DKCC) during the preverbal stage is a value-added predictor of growth in expressive language in children with autism spectrum disorder (ASD; Yoder et al., 2015). Following a theory proposed by Stoel-Gammon (2011), this study sought to determine which of four putative predictors of DKCC growth had "added value" in predicting variance in DKCC growth in initially nonverbal children with ASD. Additionally, in accordance with Stoel Gammon’s framework for phonological development, we tested whether receptive language might mediate the association of predictors with growth of DKCC. Identifying the value-added predictors of DKCC growth, and elucidating the mechanisms by which these predictors influence DKCC growth, may shed light on how we might most effectively target this factor in young children with ASD.

Methods: DKCC of eighty-seven initially nonverbal preschoolers with ASD was assessed five times over 16 months. Growth curve modeling was used to estimate initial level of, and average rate of growth in, DKCC. Putative early predictors included motor imitation, attention during child-directed speech, intentional communication and parent linguistic response to child leads. Cognitive impairment and ASD symptomatology were controlled. Simple mediation models were tested by examining whether bootstrapped confidence intervals around the indirect effect (i.e., the effect of putative predictors on DKCC through receptive vocabulary) excluded zero (Hayes, 2009).

Results: We found that children's intentional communication and parent linguistic responses to child leads predicted DKCC growth, after controlling for the two other putative predictors, as well as cognitive impairment and ASD symptomatology. As predicted, receptive vocabulary mediated the association of the value-added predictors with endpoint DKCC.
Discussion: These findings lend empirical support to the transactional theory of speech sound development. We confirmed that one parent factor (linguistic input) and two child factors (intentional communication and receptive vocabulary) contribute in a dynamic manner to impact growth in vocal communication (i.e., DKCC) in children with ASD.

References/Citations:

Paper 3 of 4

Paper Title: Does Child Vocal Contingency Predict Characteristics of Speech in Preschoolers with Autism Spectrum Disorder?

Authors: Amy Harbison¹, Paul Yoder²

Introduction: Reciprocity is essential to linguistic communication between two people. A lack of reciprocity is one diagnostic criterion for autism spectrum disorder (ASD), but there is little research about vocal responsiveness in ASD. In this study, we propose a novel measure of child vocal responsiveness: child vocal contingency (CVC), an index of the degree to which child vocalizations are contingent on adult speech, independent of base rates of both speakers' vocalizations. Even small differences in CVC could cumulatively yield vast differences in the number of opportunities for interaction and practice with responsive vocalizations. Additionally, variance in CVC might reflect the degree to which children attend and respond to adult speech, which in turn might influence the degree to which children use speechlike communication when they cannot yet use words to communicate. The purpose of this exploratory study was to investigate the following research questions: (a) To what extent is CVC stable across recordings within participants? (b) Across how many daylong sessions do we need to average CVC measures to attain criterion-level (i.e., g ≥ 0.8) stability? (c) Does CVC positively correlate with concurrent speech likeness of vocalizations (SLV)? (d) Does motor imitation moderate the relation between CVC and SLV?

Methods: We used Language ENvironment Analysis recorders to gather and code daylong audio recordings collected on two consecutive days for 33 minimally verbal preschool-age children with ASD. We used sequential analysis to derive CVC from automatically categorized vocal data. SLV was derived automatically from the same recordings. The measure of motor imitation was a composite score made up of total scores from the Motor Imitation Scale and scores from the Nonverbal Volitional Oral Abilities subscale of the Oral Motor Examination. We conducted a generalizability and decision study using each participant's audio recordings to analyze the stability of CVC across recordings within participants.

Results: Our stability criterion for CVC was surpassed within two recordings (g = 0.86) and nearly met with one recording (g = 0.76). The correlation of CVC and SLV approached, but did not meet, statistical significance (r = .349, p = .059). Motor imitation did moderate the effect of CVC on SLV. This interaction had a large effect size (R2 change = .325, p < .001). The slope of the association between CVC and SLV was significant for 74% of participants.

Discussion: The significant interaction between CVC and motor imitation in predicting SLV indicates that children with ASD who tend to respond vocally to adult speech and who imitate adult actions produce more speechlike vocalizations, which predicts
later spoken vocabulary. CVC is also a stable measure of child responsiveness. These are compelling reasons to use CVC in future research.

References/Citations:

Paper 4 of 4

Paper Title: Mother-Infant Vocal Coordination and Language Development in Infants at Heightened Risk for Autism Spectrum Disorder

Authors: Jessie Northrup2, Jana Iverson2

Introduction: Research on typically-developing infants indicates that coordination in early parent-infant vocal interactions is related to later developmental outcomes, particularly in language (Jaffe et al., 2001; Tamis-LeMonda, Bornstein, & Baumwell, 2001). Infant siblings of individuals with autism spectrum disorder (ASD) are at heightened risk (HR) for being diagnosed with ASD, as well as for significant language delays (Zwigenbaum et al., 2005). Thus, the current study was designed to examine how vocal coordination in early mother-infant interactions relates to later language abilities in HR infants.

Methods: 52 HR infants were videotaped during a 5-minute unstructured toy play interaction with their mothers when infants were 9 months of age. All mother and infant vocalizations were coded on a moment-by-moment basis, and infant vocalizations were categorized as pre-speech (PS; containing vowels and/or consonants) or non-speech (NS; e.g. unvoiced sounds, growls, raspberries). This coding generated the following variables: frequency of vocalizations, average duration of latency to respond (the time between when one person stops speaking and their partner responds) and frequency of simultaneous speech (when one speaker speaks during a partner’s vocalization).

Infants were followed longitudinally, and two standardized measures of language (Macarthur Bates CDI and Mullen Scales of Early Learning) were administered at 18, 24, and 36 months. These measures were used to classify infants as language delayed (LD, n = 15) or not delayed (ND, n = 30) and to create a continuous measure of language ability in toddlerhood. In addition, 7 infants were diagnosed with ASD at 36 months. Statistical analyses focused only on dyads with ND and LD infants, but descriptive reports of dyads with ASD infants will also be reported.

Results: While there were no differences between ND and LD infants or their mothers in the production of vocalizations, differences emerged in coordination of vocalizations. Mothers of LD infants responded faster to infant vocalizations than mothers of ND infants. However, this was driven by maternal responses to NS vocalizations. While there was no difference
between groups in responses to PS vocalizations, mothers of LD infants responded faster to NS vocalizations than mothers of ND infants. Interestingly, mothers of ASD infants showed the fastest responses to NS vocalizations of all three groups.

In addition, LD infants produced more simultaneous speech than ND infants, and this was particularly true for PS vocalizations. The percent of LD infants' PS vocalizations that were simultaneous was significantly higher than that of ND infants (and descriptively higher than that of ASD infants), and this variable was predictive of a continuous measure of language in toddlerhood.

Finally, the degree to which mothers and infants were coordinated (i.e., similar to one another) in latencies to respond was a significant predictor of later language ability.

**Discussion:** Results indicate that differences in how mother-infant dyads coordinate vocalizations in an interaction are predictive of infants' later language outcome. Findings will be discussed in terms of early markers of language delay in HR infants and the potential cascading effects of early delays on development.

**References/Citations:**

Symposium Title: Mild Cognitive Impairment and Dementia in Adults with Down Syndrome: Early Indicators of Clinical Progression

Chair: Sharon J. Krinsky-McHale

Discussant: Elisabeth Dykens

Overview: Individuals with Down syndrome are at high risk for developing Alzheimer’s disease-related dementia. The current increase in the number of aged individuals with Down syndrome warrants reliable strategies to improve early detection of cognitive impairment and to identify individuals at highest risk for developing dementia. This symposium will examine the cognitive, functional and genetic indicators that show promise for identifying clinical progression of decline.

Paper 1 of 4

Paper Title: Early Indicators of Clinical Progression of Alzheimer’s Disease in Adults with Down Syndrome

Authors: Wayne Silverman, Sharon J. Krinsky-McHale, Warren B. Zigman

Introduction: The high risk for Alzheimer’s disease (AD) experienced by middle-aged adults with Down syndrome (DS) has attracted considerable attention over recent decades. This reflects broadening recognition of the expanding population of affected adults, as well as the need to gain a greater understanding of relevant pathways in order to guide development of effective treatment and prevention. Progress toward these goals depends upon valid differentiation between individuals with DS developing AD-related changes and those who do not, and considerable attention has been focused on this issue. However, variability in lifelong abilities complicates diagnosis and as yet no consensus has emerged regarding methods for defining early stages of clinical AD progression. We have been conducting a large prospective study of older adults with DS, documenting functional, cognitive and health status. Longitudinal findings provide a rich body of data documenting characteristics of AD-related decline as well as stability in clinical status. This presentation will discuss the potential of several measures of cognition for detecting early AD-related decline in adults with DS, including mild cognitive impairment (MCI-DS), and, by extension, intellectual disability more broadly.

Methods: Individuals have been assessed at 14-22 month intervals, each cycle ending with a consensus conference classifying dementia status based on all available information. Dementia status can then be related to specific measures of cognition, including: (a) mental status (e.g., modified MMSE, Test for Severe Impairment), (b) verbal fluency, (c) block design (from the WISC + easier items), (d) visual-motor integration, and (e) episodic memory (modified Selective Reminding Test). While over 600 adults with DS have participated for up to 9 assessment cycles, the present analyses included only those individuals who did not have clinical indications of AD-related declines initially and, over the course of three years: (a) remained without MCI-DS or dementia (n = 106; mean age = 47.7 years), (b) developed MCI-DS without progressing to dementia (n = 35; mean age = 56.4), and (c) developed MCI-DS and then progressed to dementia (n = 25; mean age = 56.4). Thus, only "new" cases of MCI-DS or dementia were included, providing an explicit focus on early clinical indications of AD.

1 New York State Institute for Basic Research in Developmental Disabilities
2 Vanderbilt University, Kennedy Center
3 Kennedy Krieger Institute
4 Johns Hopkins University School of Medicine
Results: Group (3) by Time (3) multivariate repeated measures analyses of variance examined group effects. Group by Time interactions were consistently significant, with groups developing MCI-Ds declining significantly more than the group that did not. Further, the group progressing to dementia consistently showed the greatest decline over time. (A modified version of the Mini-Mental State Exam developed specifically for use with the DS population serves as an example - interaction multivariate F (4,262) = 19.4, p < .0001.) In addition, some specific measures showed cross-sectional group differences at Time 1, prior to initial development of either MCI or early dementia, suggesting sensitivity to "early MCI", a stage of AD that may be particularly responsive to intervention.

Discussion: Findings consistently showed that the methods we have developed are sensitive to early AD-related decline in adults with DS. No doubt the same is true for methods developed by other interested groups, and the field now needs a consensus on methods and criteria that provide valid determinations of dementia status for individual adults with DS. We are continuing to explore combinations of measures that provide the sensitivity and specificity needed for this task, for both MCI-Ds and early dementia, to support development of improved clinical diagnosis as well as future studies of potential biomarkers.

Acknowledgements: Supported by NIH grants P01 HD035897 and U54 HD079123 (Silverman) and funds from the New York State Office for People with Developmental Disabilities.

Paper 2 of 4

Paper Title: Gait Changes May Signal Mild Cognitive Impairment and Dementia in Adults with Down Syndrome

Authors: Sharon J. Krinsky-McHale^1, Edmund C. Jenkins^1, Joseph H. Lee^5, Nicole Schupf^5, Warren B. Zigman^1, Wayne Silverman^3,4

Introduction: Recent studies of Alzheimer’s disease (AD) in individuals in the general population have suggested that altered gait may be observed during in the earliest stages of cognitive decline, including mild cognitive impairment (MCI). It is not very clear though, which specific characteristics of gait (e.g., speed, rhythm or cadence, stride length) are linked to cognitive decline. Given the high risk of dementia in older adults with Down syndrome (DS), characterizing gait dysfunction may aid in the diagnostic process.

Methods: All participants were enrolled in a multidisciplinary longitudinal study focused on aging and dementia. Gait assessments were available for 174 individuals. During the course of the study it was determined that 50 participants (Mage=56.8; MIQ=36.7) exhibited MCI-Ds and 35 participants (Mage=61.1; MIQ=36.0) exhibited dementia, 89 participants (Mage=55.4; MIQ=37.7) remained cognitively intact.

All participants received comprehensive evaluations at approximately 14-22 month intervals. Gait was assessed using the Tinetti Gait Evaluation which quantifies gait disturbances in 7 domains by having the individual stand with the examiner and walk down a hallway or across the room, first at their "usual" pace, then back at a "rapid" pace. Following testing the dementia status of each participant was rated based upon consideration of cognitive, functional and health status data.

Results: The presence of gait dysfunction as measured by the Tinetti Evaluation varied with severity of cognitive decline in adults with Down syndrome, with 49.4% of participants who were cognitively intact, 64.0% of participants with MCI and 71.4% of participants with dementia exhibiting at least one impairment in gait (chi-square (2, 174)=6.0, p<.05). However, specific aspects of gait were differentially related to dementia severity. With increasing cognitive impairment there was an increase in hesitancy to initiate gait, observed in 34.3% of participants who were cognitively intact, 64.0% of participants with MCI and 71.4% participants with dementia (chi-square (2, 174)=10.7, p=.005); an increase in step discontinuity with 10.1% of participants who...
were cognitively intact, 16.0% of participants with MCI and 28.6% of participants with dementia (chi-square (2, 174)=6.54, p=.038); a marked deviation in path excursion with 1.1% of participants who were cognitively intact, 8.0% of participants with MCI and 17.1% of participants with dementia (chi-square (4, 174)=12.48, p=.014) and there was an observed impairment in walking stance with 36.0% of participants who were cognitively intact, 46.0% of participants with MCI and 60.0% of participants with dementia (chi-square (2, 174)=6.06, p=.048). The association between dementia severity and step length/height, step symmetry and trunk stability was not significant.

Discussion: This study linked changes in gait to the progression of cognitive impairment in adults with Down syndrome and clarified the specific characteristics of gait that may be linked to decline. While an analysis of gait will not replace a comprehensive neuropsychological assessment to diagnose an individual’s cognitive impairment, it may prove to be an important tool to inform diagnosis, track treatment effects and disease progression. Step discontinuity and deviation in path excursion, two concerns that are rarely observed in the absence of MCI or dementia, might be particularly informative when observed in an individual suspected of cognitive decline. Gait control depends on complex brain processes that involve the effective integration of motor, perceptual, and cognitive processes, AD disrupts the involved pathways. A systematic examination of gait might provide a window into another aspect of brain function during the clinical onset of dementia.

Acknowledgements: Supported by funds from the New York State Office for People with Developmental Disabilities and NIH grants P01 HD035897 and U54 HD079123 (Silverman).

Paper Title: Using Informant-Based Measures to Identify Early Indicators of Clinical Progression of Alzheimer’s Disease in Adults with Down Syndrome

Authors: Warren B. Zigman¹, Edmund C. Jenkins¹, Joseph H. Lee⁵, Nicole Schupf², Sharon J. Krinsky-McHale¹, Wayne Silverman¹⁴

Introduction: Improved understanding of the neurobiologic insults that are related to the development of Alzheimer’s disease (AD) has resulted in extensive efforts to develop treatments to forestall or ameliorate clinical deterioration. This is especially essential for adults with Down syndrome (DS), who invariably develop neuropathological hallmarks of AD and are at a high risk for dementia onset by late middle age. While there may be some encouraging progress in the neurotypical population toward attaining these goals, progress has been more limited for adults with DS. This may be due, at least to some degree, to a lack of consensus diagnostic criteria that differentiates the onset of dementia from lifelong deficits in cognitive and adaptive functioning. Until we can reliably discriminate between individuals with DS who develop AD-related clinical changes and those who do not, progress toward prevention or treatment will be seriously hindered. We have been following a large sample of adults with DS for many years, documenting functional, cognitive and health status with the goal of reliably classifying the various stages of AD (i.e., non-demented, mild cognitive impairment (MCI) and frank dementia). This presentation will discuss the potential of several measures provided through informant interviews for the early detection of AD-related decline.

Methods: Over 600 adults with DS have participated for as many as 9 cycles of data collection over the past 15 years. Functional, cognitive and health status has been documented at 14-22 month intervals, with each cycle resulting in a classification of dementia status based on all available information. The present analysis was designed to relate dementia status to two measures of cognitive and functional behavior, the Dementia Questionnaire for People with Learning Disabilities (DLD) and the Adaptive Behavior Scale (ABS Part I). Evenhuis developed the DLD as an informant-based questionnaire, to measure cognitive and functional deterioration that occurs because of dementia. The DLD provides two summary scores, a Sum of Cognitive Scores (SCS) and a Sum of Social Scores (SOS). The ABS Part I was designed to measure basic activities of daily living and instrumental activities of daily living in intellectually and/or developmentally disabled individuals. The present analyses included only those
individuals who did not have clinical indications of AD-related declines prior to enrollment and, over the following three years: (a) remained without MCI or dementia (n = 38; mean age = 54.98 years), (b) developed MCI without progressing to dementia (n = 53; mean age = 54.27), and (c) developed MCI and then progressed to dementia (n = 32; mean age = 54.21). Mean age [F (2,122) < 1, NS] and sex composition [Chi-Square (2) = 1.1, NS] did not differ among groups.

Results: Group (3) by Time (3) multivariate repeated measures analyses of variance examined group effects. Group by Time interactions were consistently significant [all Fs (4,240) > 9, ps < .001], demonstrating that the group progressing to dementia consistently showed the greatest decline over time and the group developing MCI declined a lessor, but more substantial degree than the group that remained non-demented.

Discussion: These findings, in conjunction with those reported by Silverman et al. (this symposium), indicate that informant-based and individual cognitive testing both provide indications of early AD-related decline for adults with DS. The ability to assess dementia status with minimal participant interaction may allow broader screening of large populations of adults with DS and perhaps intellectual disabilities due to other causes, enabling early interventions, once developed, to mitigate loss of function.

Acknowledgements: Supported by funds from the New York State Office for People with Developmental Disabilities and NIH grants, R01 AG014673 (Schupf), P01 HD035897 and US4 HD079123 (Silverman).

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Paper 4 of 4

Paper Title: SnpS in CUGBP2 Influence the Risk of Alzheimer’s Disease in Adults with Down Syndrome

Authors: Joseph H. Lee, Rong Cheng, Lam-ha Dang, Sharon J. Krinsky-Mchale, Warren B. Zigman, Wayne Silverman, Benjamin Tycko, Lorraine Clark, Nicole Schupf

Introduction: The CUGBP2 gene, also known as CELF2, was first reported to be associated with familial late-onset Alzheimer disease (LOAD) in White and Caribbean Hispanics, especially in the presence of homozygous APOE ε4, suggesting that CUGBP2 may be involved in regulation or processing of amyloid β (Aβ) peptides. In addition, a meta-GWAS of an international consortium dataset identified CELF1, a homolog of CUGBP2/CELFI2, as a risk gene for Alzheimer disease (AD). Since individuals with Down syndrome (DS) are exposed to excess levels of Aβ peptides from having three copies of the APP gene, we examined CELF1 and CUGBP2 genotypes and dementia risk for a cohort of adults with Down syndrome (DS). Subsequently, we examined a larger set of Caribbean Hispanic and white non-Hispanic cohorts as well as the Alzheimer Disease Neuroimaging Initiative (ADNI) cohort.

Methods: We first examined 320 adults with DS who participated in a candidate gene study to assess risks associated with AD clinical progression and levels of Aβ. We then examined multiple datasets comprising two different ethnic groups to evaluate further, the allelic association between CUGBP2 and AD. Specifically, we performed meta-analysis using 1,961 genotyped or high-quality imputed SNPs. After combining Caribbean Hispanic elderly and White participants, totaling 6,509 individuals, we applied logistic regression to estimate the risk of AD, adjusting for age, sex, APOE ε4, and population structure. In addition, we performed multivariable survival analysis to assess the relationship between SNPs and age at onset/last examination, adjusting for the same confounders.

Results: We evaluated allelic association using SNPs that overlap in the cohorts of adults with DS and those without DS. We found one SNP, rs2378991, that was associated with dementia as well as age at onset (p=0.04) in both adults with DS and adults without DS. In addition, several SNPs were associated with AD in Hispanic and white elderly; however, we were unable to test some of the SNPs in adults with DS because the number of SNPs in DS was far fewer than that in the general population. We

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6 Columbia University School of Medicine
note that the meta-analysis of Hispanics and whites identified four SNPs for age at onset of AD (rs10905930, rs12268544, rs3847422, and rs2378991), located with a 1.8kb region, with p-values ranging from 2x10⁻⁴ to 9x10⁻⁴. In addition, rs10795847 was associated with elevated risk of AD (p=9x10⁻⁴).

**Discussion:** Our multi-stage study showed that genetic factors in the CUGBP2 gene are likely to be involved in AD, and may work through the amyloid pathway in both adults with and without DS. Additional siRNA experiments are ongoing to further probe the functional relevance of this gene.

**Acknowledgments:** Supported by grants R01AG014673 (Schupf) and P01HD35897 and US4 HD079123 (Silverman) from NIA and NICHD and by NYS through its Office for People with Developmental Disabilities.
**Symposium Title**: Parent Well-Being of Youth with Developmental Disabilities: Implications and Interventions

**Chair**: Elizabeth Halstead¹ ²

**Discussant**: Richard P. Hastings¹ ³

**Overview**: This symposium addresses a gap in current IDD literature exploring factors or processes to be targeted in intervention and intervention evaluations surrounding parental well-being of youth with developmental disabilities. Two research types are included in this symposium; including studies which are focused on generating information to inform interventions, and studies which are focused on intervention evaluation. The two studies which focus on implications for intervention explore child behavioural difficulties measured by maternal reporting, and mothers’ well-being. Resilience and associated mental disorders were explored as factors that may influence this relationship. The first intervention study involves a randomized controlled trial targeting child emotion regulation, comparing treatment to a wait-list control group. Parental mental health was also assessed as a factor relating to well-being and child emotional regulation. The second intervention study is a pilot study aimed to evaluate the effectiveness of a therapeutic education intervention for parents of children with autism, this includes a multidimensional approach to promote interaction and group dynamics. This symposium brings together different factors or processes relating to maternal well-being and provides clear direction for further intervention development for families of children with IDD.

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**Paper 1 of 4**

**Paper Title**: Longitudinal look at psychiatric disorder and family impact in adolescents with ASD: Implications for intervention

**Authors**: Jan Blacher⁴ ⁵, Amanda Norona⁵, Bruce L. Baker⁵

**Introduction**: Children and adolescents with Autism Spectrum Disorder (ASD) have higher rates of behavior problems and mental disorders than their peers with typical cognitive development (TD) (Totsika et al., 2011). We have found the rate of diagnosed ADHD to be over 3 times as high in ASD than in TD youth (Baker & Blacher, 2015). A concomitant finding has been that parents of youth with ASD and behavior disorders have considerably heightened stress and poorer psychological adjustment. In the present study, we examined these relationships further, by a longitudinal analysis of youth behavior problems and maternal well-being at two points in early adolescence (ages 13 to 15). These data were drawn from the Collaborative Family Study, a 15-year longitudinal (3 to 15 years) project focusing on families of children with Intellectual Disability (ID), ASD, and TD.

**Methods**: Participants were mothers and adolescents, assessed at youth ages 13 and 15 years. Youth had typical cognitive development (n = 88) or ASD (n = 47). While the TD youth had IQs of 85 or higher (WISC), the ASD group youth was comprised of youth across the IQ spectrum, i.e., TD and ID. A prior study (Baker & Blacher, 2015) of this sample found no relationship between youth behavior problems/mental disorders and IQ in the ASD sample.

Mothers and youth completed center-based assessments and measure packets at both assessment ages. Measures of primary interest here are: (1) Child Behavior Checklist (CBCL), externalizing and internalizing broadband scales; ADHD, ODD, and Anxiety clinical scales; (2) Family Impact Questionnaire (FIQ), a parent-report measure of how the respondent views the child’s impact on

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⁴ Graduate School of Education, University of California Riverside
⁵ Department of Psychology, University of California, Los Angeles
the family; Negative Impact scale (a proxy for parenting stress) and Positive Impact scale; (3) Mother's Health Report, a self-report on a single four-point scale (Excellent, Good, Fair, Poor) that has strong statistical properties despite its brevity.

We addressed three primary research questions: (1) Are behavior problems/mental disorders stable across early adolescence for youth with TD or ASD? (2) Does parent well-being change across the youth's early adolescence? and, especially, (3) Do changes in parent well-being relate to changes in youth behavior problems/mental disorders?

**Results:** On every behavior disorder assessed, youth with ASD scored higher than youth with TD. This is consistent with earlier findings of the considerably increased psychiatric difficulties of youth with ASD. An unexpected finding was that on every variable assessed, youth difficulties decreased significantly from age 13 to 15 year. While mothers' reports of positive impact remained stable, mothers' stress and health problems also decreased significantly during this early adolescent period. Finally, the decrease in youth disruptive behavior and the decrease in mothers' parenting stress were positively correlated, within both the TD (r = .58, p <.001) and ASD (r = .31, p <.05) groups.

**Discussion:** We will conduct further analyses on this correlated downward trajectory of youth behavior disorders and mother stress in early adolescence, and discuss implications for intervention in the relatively neglected period of adolescence. This finding could also enhance earlier intervention efforts by instilling optimism and hope early on in parents of children with ASD who struggle with their child's disruptive behavior disorders.

**References/Citations:**


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**Paper 2 of 4**

**Paper Title:** Does Resilience Function As A Protective or Compensatory Factor for the Well-Being of Mothers of Children with Developmental Disabilities?

**Authors:** Elizabeth Halstead¹², Gemma M. Griffith², Richard P. Hastings¹³

**Introduction:** Behaviour problems exhibited by children with intellectual and developmental disabilities (IDD) have been identified as a significant stressor for family members in both cross-sectional and more recently longitudinal research (Lecavalier et al., 2006; Zeedyk & Blacher, 2015). Despite the consistency of the relationship between child behaviour problems and maternal well-being in existing research, there is variability in mothers' responses to their child's behaviour problems. Not all mothers whose child has significant behaviour problems reports increased psychological distress or lower levels of well-being (Hill & Rose, 2009). In theoretical terms, this suggests that there are some factors that mediate or moderate the impact of child behaviour problems on mothers' well-being. One additional concept that has been used in IDD family research to explain some of the variation in maternal well-being is that of resilience. In the present study, we explored whether maternal resilience explains some of this variability and specifically whether resilience functions as a protective or compensatory factor.

**Methods:** Participants were 312 mothers of children with intellectual and developmental disabilities (IDD) and aged between 4 and 15 years old (M= 10.02, SD =3.08). Mothers were asked to participate in a cross-sectional online survey.
Measures included: Parent and Family problems subscale from the Questionnaire on Resources and Stress - short form (QRSF7, Griffith et al., 2011); The Positive Gain Scale (MacDonald et al., 2010); Pit-ten Cate, 2003); The Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983); Family satisfaction was measured by the Family Satisfaction Scale (Olson & Wilson, 1982), The Brief Resilience Coping Scale (Sinclair and Wallston, 2004) is designed to assess an adult’s ability to bounce back from stress.

Results: Using moderated multiple regression models, we found consistent evidence that maternal resilience functioned as a compensatory factor - having a significant independent main effect relationship with maternal stress (R = .611, R2 = .374, F= 19.480, p = .004), anxiety (R=.502, R2 = .252, F= 10.590, p<.001), depression (R= .524, R2 = .275, F= 11.203, p<.001), perceptions of positive gain (R=.394, R2 = .155, F= 5.305, p<.001) and family satisfaction R=.535, R2 = .286, F= 12.645, p<.001). There was little evidence of the role of resilience as a protective factor between child behaviour problems and maternal well-being.

Discussion: This study found that resilience as a moderator is significant only between child behavioural problems and maternal stress. In all five outcome models resilience is shown as a main compensatory effect and therefore showing the function of resilience may vary by outcome. However, it is more likely that resilience is clearly shown as having a consistent compensatory function between child behavioural problems and maternal outcomes. Building maternal resilience may result in improved overall well-being for mothers of children with IDD, and resilience over time should be explored in future research.

References/Citations:
Methods: Participants included 34 children with ASD (94.1% male), 8-12 years of age (M = 9.50, SD = 1.13) with at least average IQ (M = 103.48, SD = 14.06), and their caregivers (76.5% mothers), involved in a randomized controlled trial targeting child emotion regulation, comparing treatment to a wait-list control group. Parent mental health was assessed using the Depression Anxiety & Stress Scale-21 (Lovibond & Lovibond, 1995), and emotion regulation by the Cognitive Emotion Regulation Questionnaire-Short Version (Garnefski & Kraaij, 2006). Child psychopathology was assessed via parent report on the Behavior Assessment System for Children, Second Edition (Reynolds & Kamphaus, 2004), and independent clinician judgment using the Clinical Global Impression Scale - Improvement (Guy, 1976).

Results: Post-intervention, there was a significant reduction in parent depression, t(31) = 2.10 p = .04, and severity of overall parent distress, t(31) = 2.27, p = .03 from pre-intervention levels. Controlling for baseline scores, parents in the treatment group improved relative to the waitlist group on acceptance, F(1,30) = 4.36, p = .045, catastrophizing, F(1,30) = 5.37, p = .03, depression, F(1,29) = 5.40, p = .03, stress, F(1,30) = 4.96, p = .03, and total distress, F(1,29) = 6.38, p = .02. Parent improvement in catastrophizing was correlated with improvement in parent-reported child internalizing symptoms (r = .42, p = .02), and parent reduction in anxiety was related to overall child clinical improvement according to clinician judgment (r = .47, p = .03). Contrary to expectation, parent improvement in refocusing was associated with less improvement in parent-reported child externalizing symptoms (r = -.37, p = .04).

Discussion: Findings will be discussed in relation to optimally involving parents in therapy for children with ASD to promote positive parent and child outcomes.

References/Citations:
Introduction: Autism Spectrum Disorders (ASD) have major implications for the individual functioning but also on the family environment, especially on parents. As highlighted by the last French Autism Plan (French Health and Social Ministry, 2013), it is crucial to develop and evaluate programs for parents in order to support several dimension of parenting (skills, knowledge, emotional adjustment). The ETAP program is based on a multidimensional approach and was developed from a preliminary study about specific needs of parents of children with ASD (Derguy, Michel, M'Bailara, Roux, & Bouvard, 2015). It consists of seven structured group sessions of 1:30 over a four months period. Educational techniques and structured activities were used in order to promote interaction and group dynamics. The topics covered include: (1) "What is autism for me? "; (2) The communication with my child; (3) The relationship between the behaviour and emotions of my child; (4) Social isolation and stigmatization; (5) Relationships with my family and my environment; (6) Interventions for my child; (7) Assessment of my participation to the ETAP program. This pilot study aimed to evaluate the effectiveness of a therapeutic education intervention for parents of children with autism.

Methods: Participants included 40 parents (82.5% mothers) of children with ASD, 3-10 years of age divided in two groups (ETAP versus control group). The ETAP group included 30 parents (mean age: 39.3 ± 6.7 [25 -60]), which participated in all sessions. The wait-list control group included 10 parents (mean age: 38 ± 3.4 [34-45]).

To assess the impact of the program, we conducted measurements for both groups before the intervention (T1) and 4 months later, after the intervention (T2).

Parents quality of life was assessed using the French version of the WHOQOL-BREF (Baumann, Erpelding, Régat, Collin, & Briançon, 2010; The WHOQOL Group, 1995). Parents depressive symptoms were assessed using the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983).

Results: After the program, there were significant improvements in quality of life (U=79; p=.033) and a significant reduction of depressive symptoms (U=80.5 ; p=.036) only for parents who have participated to the ETAP program.

Discussion: This first session provides encouraging results about the social validity (attendance, satisfaction regarding the content and the organization, etc.) and the benefits of a therapeutic education intervention to understand and support parental adjustment in autism spectrum disorders.

References/Citations:
Symposium Title: Age Related Psychological Phenotypes in Genetic Disorders

Chair: Chris Oliver

Overview: An emerging theme in behavioral phenotype research is the change in cognitive, behavioral and social phenotypes that is associated with early and later ageing. Both cross-sectional and longitudinal designs have the capacity to identify atypical developmental trajectories within and between syndromes across psychological domains. In this symposium cognitive, social and behavioural phenotypes in a number of genetic disorders are examined against the background of early and later ageing.

Paper 1 of 4

Paper Title: Early Predictors of Adult Quality of Life for Individuals with Neurogenetic Syndromes

Authors: Jess Penhallow\textsuperscript{1}, Jo Moss\textsuperscript{1,2}, Henna Ahmed\textsuperscript{1}, Chris Oliver\textsuperscript{1}

Introduction: A major concern of parents whose child has been diagnosed with a genetic syndrome is for their long term development and future life opportunities. However, for the majority of these rare syndromes lifespan research is scarce. This may leave parents with an uncertain outlook for their child’s future.

Long term outcome measures of Quality of Life (QoL), may help to address some of the concerns parents have over their child's future. This study aims to describe and compare the QoL of individuals with five genetic syndromes and to utilise longitudinal date to explore early predictors of adult QoL.

Methods: Parents and carers of individuals with Angelman syndrome (AS; N =7), Cri du Chat syndrome (CdCs; N= 6), Cornelia de Lange syndrome (CdLS; N=13), Fragile X syndrome (FXS; N=20) and Prader-Willi syndrome (PWS, N=13) completed a variety of questionnaires describing the behaviour of the person they cared for. These included measures of adaptive behaviour (The Wessex Questionnaire, Kushlick et al., 1973), challenging behaviour (Hyman et al., 2002), characteristics of autism spectrum disorder (ASD; the Social Communication Questionnaire; Rutter et al., 2003), overactivity and impulsivity (The Activity Questionnaire; Burbidge & Oliver, 2008) and mood interest and pleasure (MIPQ; Ross et al, 2008).

After a follow-up period of 12 years they completed the World Health Organization Quality of Life scale (WHOQOL-Bref; World Health Organization, 1996) including an additional module that addresses factors that specifically relate to the quality of life of individuals with an intellectual disability (WHOQOL-DIS; Power & Green, 2011). The WHOQOL-Bref defines QoL in four domains of: physical health related QoL, psychological QoL, social QoL and environmental QoL.

Results: The participants’ quality of life was significantly lower than population norms in all domains except for the environment domain where their scores did not significantly differ from population norms \([t (921) = .97, p = .33]\). Scores on the environmental QoL domain were significantly higher than those in the other three domains of physical health related QoL, psychological QoL and social QoL \([F(2.11, 107.49) = 18.99, p<.01]\). However, when compared to a population of individuals with intellectual disability of heterogeneous cause on the WHOQOL-DIS module, our sample’s QoL scores were significantly higher \([t (1187) = 14.72, p<.01]\). There were no significant differences between the five syndrome groups in any WHOQOL-BREF domain scores or in WHOQOL-DIS scores.

There were several significant relationships between aspects of the participants' early behavioural phenotypes and their later quality of life. Levels of mood interest and pleasure and the number of ASD characteristics displayed by the participants were

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significantly related to scores on the WHOQOL-DIS ($r = .29, p < .01; r = -.36, p < .01$) and a number of domains on the WHOQOL-BREF.

**Discussion:** This is the first study to explore the predictors of QoL in adults and adolescents with genetic syndromes. From our findings it appears that individuals with different genetic syndromes do not significantly differ from each other in their QoL outcomes. These outcomes are poorer than those of the general population but are comparable to, and possibly more positive than, those of others with an intellectual disability. Low mood interest and pleasure and the presence of characteristics associated with ASD may predict poorer quality of life outcomes.

**References/Citations:**

**Paper 2 of 4**

**Paper Title:** The Correlates of Self-Injurious Behaviour and Aggression in Lowe Syndrome: A Longitudinal Study

**Authors:** Jane Waite¹, Alicia Kutsch¹, Lucy Wilde¹, Kate Eden³, Chris Stinton⁴, Jo Moss¹ ², Chris Oliver¹

**Introduction:** Self-injurious and aggressive behaviours occur in 60-70% of individuals with Lowe syndrome in comparison to between 10-15% of individuals with intellectual disability of heterogeneous aetiology, therefore, further examination of the correlates of behaviour disorder in Lowe syndrome is of interest. In cross sectional studies, overactivity, impulsivity and stereotyped behaviours have been associated with self-injurious behaviours, and compulsive behaviour and impulsivity have been associated with aggressive behaviours (Arron et al., 2011). To date, there have been no longitudinal follow-up studies of behaviour in Lowe syndrome. This study explores how the correlates of challenging behaviour have changed over eight years in those who exhibit aggression and self-injurious behaviour.

**Methods:** Longitudinal questionnaire data were collected on twenty-one individuals with Lowe syndrome with a mean age of 16.0 years (range: 4.0-34.9). Participants were recruited through the Lowe syndrome Trust (UK) and Lowe Syndrome Association (USA). Parents/carers completed a questionnaire pack at baseline (T1) and at eight year follow-up (T2). Measures included the Activity Questionnaire, the Repetitive Behaviour Questionnaire and the Challenging Behaviour Questionnaire. Degree of ability was measured using the Wessex Scale. A series of Mixed ANOVAs with one between group factor (SIB at T2) and one within factor (time) were employed to explore differences in correlates of self-injurious behaviour at each time point. These analyses were repeated for aggression. Post-hoc comparisons were conducted. A p-value of $p < .05$ was adopted for mixed ANOVAs and $p < .01$ for post-hoc comparisons.

**Results:** There were no differences between the SIB ($N = 10$) and non-SIB groups in age at T1 or T2. The SIB group showed significantly lower ability scores, $t(19) = 3.58, p = .002$, but did not differ on any of the other variables at T1. At T2, The SIB group had significantly higher scores on impulsivity, over-activity in comparison to the non-SIB group, $t(19) = -3.05, p = .007; t(19) = -4.48, p < .001$. Repeated measures t-tests revealed that the non-SIB group showed a significant decrease in over-activity scores over time, $t(9) = 2.95, p = .008$. Decreases in impulsivity, compulsive and stereotyped behaviour scores approached significance

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in the non-SIB group (ps .039, .033 & .033 respectively). The opposite trend was observed in the SIB group with increases in over-activity and compulsive behaviour approaching significance (ps = .017). There were no differences between those displaying (N = 11) and not displaying aggression at T2 on any variables.

Discussion: This is the first longitudinal study of how the correlates of SIB and aggression in Lowe syndrome change over time and demonstrates significant decreases in overactivity in the non-SIB group, along with a negative trend for impulsivity and repetitive behaviour. This is in contrast to stable or increasing levels of these correlates in the SIB group. The results will be discussed in relation to Oliver and Richards’ (2015) model of the development and persistence of challenging behaviour which argues that behaviours that come into an individual’s repertoire against a background of behavioural dysregulation are more likely to persist.

References/Citations:

Paper 3 of 4

Paper Title: The Cognitive Developmental Profile Associated with Fragile X Syndrome: A Longitudinal Investigation of Cognitive Strengths and Weaknesses through Childhood and Adolescence

Authors: Eve-Marie Quintin5, Jo Booil6, Scott S. Hall6, Jennifer Bruno6, Lynsey C. Chromic6, Mira M. Raman6, Amy A. Lightbody6, Arianna Martin6, Allan L. Reiss6

Introduction: Few studies have investigated developmental strengths and weaknesses within the cognitive profile of children and adolescents with fragile X syndrome (FXS), a single-gene cause of inherited intellectual impairment.

Methods: With a prospective longitudinal design and using normalized raw scores (Z-scores) to circumvent floor effects, we measured cognitive functioning of 184 children and adolescents with FXS (ages 6 to 16) using the Wechsler Scale of Intelligence for Children (WISC-III) on one to three occasions for each participant. Participants with FXS received lower raw scores relative to the WISC-III normative sample across the developmental period.

Results: Verbal Comprehension, Perceptual Organization, and Processing Speed Z-scores were marked by a widening gap from the normative sample, while Freedom from Distractibility Z-scores showed a narrowing gap. Key findings include a relative strength for verbal skills in comparison with visuospatial-constructive skills arising in adolescence and a discrepancy between working memory (weakness) and processing speed (strength) in childhood that diminishes in adolescence.

Discussion: Results suggest that the cognitive profile associated with FXS develops dynamically from childhood to adolescence. Findings are discussed within the context of aberrant brain morphology in childhood and maturation in adolescence. We argue that assessing disorder specific cognitive developmental profiles will benefit future disorder-specific treatment research. Standardization data from the Wechsler Intelligence Scale, Third Edition (WISC-III). Copyright © 1990 NCS Pearson, Inc. Used with permission. All rights reserved.

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References/Citations:


Paper Title: The Developmental Trajectory of Early Social Cognition Skills in Children with Fragile X Syndrome

Authors: Katherine Ellis¹, Chrisi Stefanidou¹, Laurie Powis¹, Ian Apperly¹, Jo Moss¹ ², Chris Oliver¹

Introduction: Individuals with Fragile X syndrome (FXS) are more likely to meet cut-off scores on measures of autism spectrum disorders (ASD), yet fine-grained analysis of ASD-like characteristics suggests these individuals present a unique socio-behavioural profile from those with idiopathic ASD and those with other genetic syndromes. Understanding why and how these socio-behavioural differences emerge requires detailed investigation into mechanisms thought to influence social behaviour in both typical and atypical populations, such as social cognition. Guttman analysis, a stringent and conservative method of scaling, indicates that a wide range of social cognitive concepts such as early understanding of others intentionality (Powis, 2014) and later ToM concepts (Wellman & Liu, 2004) reliably emerge cumulatively and in a strict developmental order. Yet children with atypical social profiles, such as those with idiopathic ASD and those with Rubinstein-Taybi syndrome (RTS), have revealed unique profiles of strengths and weaknesses diverging from typical development (TD). The current study aimed to further this novel approach of outlining and exploring the influence of atypical social cognitive development on socio-behavioural phenotypes by investigating the developmental sequence of early social cognition skills within children with FXS.

Methods: Twenty-two children with FXS aged two to twelve years old (Mage=5.45, SD=2.61) participated in a cognitive assessment suitable for their age and level of ability (either the 'Mullen Scales of Early Learning' or the 'British Ability Scales-III') and the 'Early Social Cognition Scale' (Powis, 2014), a battery of six tasks assessing different abilities required to understand others intentions. Different types of intention understanding are considered ‘precursor’ abilities to fully fledged ToM abilities, and emerge in a strict developmental order in TD infants aged between 14-34 months. In order of typical attainment, tasks included: ‘helping’ (typically emerging around 14 months of age), 'Re-enactment of Intended Acts' (REI) and 'Point' (18 months), and 'Gaze' and 'Tubes-with-handles' (24 months).

Results: The group’s overall cognitive ability ranged between 15-80 months (mean=33.09, SD=14.66). The percentages of the number of children with FXS who passed each task revealed an alternative sequence to what is consistently observed in TD infants: 86% passed 'REI', 82% passed 'helping', 36% passed 'tubes-with-handles', 32% passed 'point', 23% passed 'gaze' and 18% passed 'trampoline'. McNemar's tests with Yate's correction for continuity were conducted between the five task pairs that differed in increasing difficulty. Bonferroni corrections were used to control for family-wise error. The only pair that significantly differed in difficulty were the 'helping' and 'tubes-with-handles' tasks (p=0.03); there were no significant differences in performance between 'REI' and 'helping', 'tubes-with-handles' and 'point', 'point' and 'gaze', or 'gaze' and 'trampoline'.

Discussion: This is the first study investigating the atypical developmental trajectory of social cognitive abilities in children with FXS. Results show that although these children are able to acquire very early abilities ('helping' and 'REI'); there is evidence of an overall deficit in later abilities despite many of these children reaching the developmental age that these abilities emerge. This suggests that there may be an underlying mechanism unrelated to general cognitive ability possibly disrupting social cognitive
development in children with FXS. These results will be discussed in relation to 1) previously defined trajectories in both typical infants and those with RTS, and 2) the potential underlying variables both specific and non-specific to the FXS phenotype that may be associated or implicated in the development of these later abilities such as executive function, social motivation and joint attention.

References/Citations:

**Syposium Title:** Parenting Children with ID or ASD: Behavioral, Mental Health, and Social Outcomes

**Chair:** Bruce Baker

**Discussant:** Keith Crnic

**Overview:** Children with developmental disabilities exhibit hallmark behavioral and cognitive symptoms, which typically serve as the basis for their diagnosis. The behaviors and social skills presented by these children both affect and are affected by parenting behaviors and parental well-being. The four papers in this symposium consider relationships between parenting behaviors, parent and child mental health, and child social and behavioral outcomes. The first paper examines the roles that negative parenting behaviors and parental mental health play in the development of child internalizing symptoms in early childhood among children with intellectual disability (ID) or of typical development. The second paper investigates child emotion dysregulation and cognitive ability in children with or without ID as predictors of changes in parent-child conflict across early- to middle-childhood. The third paper identifies different clusters of mothers based on their depressive symptoms to determine whether mothers exhibit stability or change in symptoms over a ten-year period and whether child characteristics (e.g., disability status) relate to cluster membership. The final paper considers the efficacy and feasibility of a parent-assisted social skills intervention in addressing social skills deficits and autism spectrum disorder (ASD) symptomatology in families of young children with ASD. Collectively, across the four presentations, we aim to expand the discussion about the roles that parents and their children with developmental disabilities play in influencing one another.

**Paper 1 of 4**

**Paper Title:** Unsupportive Parenting and Parental Mental Health in Children with or without Intellectual Disability: The Effects On Internalizing Behavior Problems

**Authors:** Naomi Rodas, Sasha M. Zeedyk, Bruce L. Baker

**Introduction:** Researchers have shown unsupportive parental reactions to be associated with higher levels of emotion dysregulation in TD children (Fabes, Leonard, Kupanoff, & Martin, 2001; Shaffer, Suveg, Thomassin, Bradbury, 2012). Furthermore, children whose parents react in a non-supportive manner when negative emotions are expressed show higher levels of inhibition and are more likely to exhibit internalizing behavior problems (Denham, Bassett, & Wyatt, 2007; Rubin, Burgess, & Hastings, 2002; Hastings & De, 2008). Previous research has demonstrated the link between poor parental mental health and child emotion dysregulation (Hoffman, Crnic, & Baker, 2006; Shaffer et al., 2012). While there has been a focus on externalizing disorders in children with ID, much is still to be learned about internalizing disorders. The present study utilized observational measures of mother and father negative parenting as well as self-report measures of unsupportive parenting practices in order to understand various aspects of negative parenting. We examined the effect of negative parenting and parental depression at child age 4 on the development of child internalizing behavior problems at child age 5 in children with or without ID.

**Methods:** This study uses data obtained from 204 mothers and 173 fathers and their children with or without ID. Data were collected at child ages 4 and 5. Measures include the CBCL (Achenbach, 2000), the Coping with Children's Negative Emotion Scale (CCNES; Fabes, et al., 1990), and the Parent Child Interaction Rating Scale (PCIRS; Belsky, et al., 1995).

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2 Arizona State University
3 University of California, Los Angeles
4 University of California, San Diego
Results: Maternal depression was positively associated with child internalizing behavior problems, as indicated by a significant unstandardized regression coefficient (B = .27, t = 3.94, p < .001). Maternal depression was also positively associated with unsupportive parenting (B=.01, t= 2.13, p<.05). For fathers, the results indicated that the cross-product term between unsupportive parenting and paternal depression on child internalizing behaviors was significant (B = 6.80, t = 2.54, p < .01). Follow-up analyses of the moderation indicated that unsupportive parenting was significantly associated with child internalizing behavior problems at clinical (conditional effect = 6.47, t = 2.67, p < .01), but not at sub-clinical levels (conditional effect = -.33, t = -.28, p = .78), of paternal depression.

Discussion: Overall, we found similar relationships between parental depression and child internalizing problems for both mothers and fathers. However, we did found differences in self-reported and observed negative parenting practices when comparing mothers and fathers. Therefore, our findings suggest that we should continue to study both mothers and fathers, as their impact is not always the same.

References/Citations:

Paper 2 of 4

Paper Title: The Interactive Effects of Child Emotion Dysregulation and Cognitive Ability on Parent-Child Conflict across Early-To Middle-Childhood

Authors: Willa A. Marquis¹, Amanda N. Norona¹, Bruce L. Baker⁴

Introduction: Though previous findings have established a link between parent-child conflict and youth mental health and social adjustment, few studies (e.g., Eisenberg et al., 2008) have examined how parent-child conflict relates to emotion regulation. No studies to our knowledge have explored this relationship early in development, where understanding predictors of change in conflict could be especially salient for intervention (Smetana, 2008). Clarifying this underlying mechanism is particularly important for children with developmental delays, who are at a heightened risk for developing psychopathology (Baker et al., 2002). In the present study, we examined the interrelation of parent-child conflict, child emotion dysregulation, and child cognitive ability and explored how emotion dysregulation and cognitive ability predict change in parent-child conflict across early- to middle-childhood (ages 3 to 7).
Methods: Participants (N = 211) were from a longitudinal study of the development of psychopathology in children with developmental delays (N = 73) and typically developing children (N = 138). Level of parent-child conflict was derived from naturalistic home observations of family interactions, while emotion dysregulation was measured using the Child Behavior Checklist - Emotion Dysregulation Index (adapted from Samson et al., 2014). Cognitive ability was measured using a continuous IQ score assessed at age 5 years. Pearson correlations were conducted to examine interrelations of primary constructs, and we used PROCESS to examine the conditional interactive effects of child emotion dysregulation and IQ on change in parent-child conflict from child ages 3 to 5 and ages 5 to 7 years.

Results: Higher emotion dysregulation was associated with lower cognitive ability across ages (r = -.24 to -.23) and with higher levels of parent-child conflict at all ages (r = .16 to .19) except the first observation at child age 3. Conflict was higher among children with lower IQ at age 5 (r = -.21) and was also higher, unexpectedly, among children with higher IQ at age 3 (r = .14). Parent-child conflict increased from ages 3 to 5 only for those children with both high emotion dysregulation and low IQ (t = 2.73, p < .01; B[SE] interaction = -.03[.01], p = .04). From ages 5 to 7, conflict increased for children who had high emotion dysregulation and either low (t = 2.34, p = .02) or moderate IQ (t = 2.01, p < .05; B[SE] interaction = -.02[.01], p = .04).

Discussion: We considered the results of our correlations in the context of parents' expectations of their child’s behavior across childhood, such that conflict may be more closely linked to dysregulation in later years when dysregulated behaviors (e.g., tantrums) are less peer-normative. We conceptualized our finding that conflict increased for children with high dysregulation and low IQ as indicative of the additive risk of cognitive delay and poor regulation in predicting family functioning across the developmental span. Finally, we also explored our results within a resilience framework and discussed implications for early intervention. From a strengths perspective, fostering emotion regulation skills early in development may be protective for family functioning in the presence of developmental vulnerability.

References/Citations:

Paper 3 of 4

Paper Title: Maternal Depressive Symptoms among Mothers of Children with or without ID: Investigating Longitudinal Stability and Transition

Authors: Sasha M. Zeedyk⁵, George A. Marcoulides⁶, Jan Blacher⁴

Introduction: Few studies exist in the ID literature tracing maternal depression longitudinally. Of those published, most are limited to periods in either early childhood or adolescence. Moreover, the extant literature that examines maternal depression longitudinally has shown discrepancies, with some samples demonstrating changing symptoms over time (e.g., Glidden & Schoolcraft, 2003) and others showing stability (e.g., Carter et al., 2009). To our knowledge, latent transition analysis has not been applied to investigate depressive symptoms in samples that include mothers of children with or without ID. The present

⁵ University of California, Santa Barbara
study aimed to identify different groups of mothers based on their depressive symptoms, to investigate whether mothers exhibited stability or change in symptoms over time, and to determine if predictors of changes in group membership could be identified.

**Methods:** Preliminary analysis fitting a growth model to the maternal depressive symptom data indicated that, on average, mothers exhibited fairly stable levels of depressive symptoms (i.e., the slope estimate was not significant). However, the variance term for the slope of depressive symptoms indicated that there was significant variability between mothers. Further, a plot of predicted ordinary least squares depression trajectories showed a pattern indicating 3-4 distinct groups of mothers (i.e., variability was also present in visual analysis of the plot).

**Results:** Preliminary analysis fitting a growth model to the maternal depressive symptom data indicated that, on average, mothers exhibited fairly stable levels of depressive symptoms (i.e., the slope estimate was not significant). However, the variance term for the slope of depressive symptoms indicated that there was significant variability between mothers. Further, a plot of predicted ordinary least squares depression trajectories showed a pattern indicating 3-4 distinct groups of mothers (i.e., variability was also present in visual analysis of the plot).

A latent class analysis was then utilized to group mothers at each time point. Model fit using the Bayesian Information Criterion index indicated that a 4-class model was the best fit to the data. The results indicated that mothers fell into either low, borderline, high or very high symptom groups. The number of mothers falling into each group remained fairly stable over time. While the majority of mothers fell into the low or borderline groups (i.e., not meeting the clinical cutoff of 16 on the CES-D), 11-17% (depending on the time point measured) fell into the high or very high groups, displaying symptoms well above the clinical cutoff. Additional analyses will be conducted to investigate whether mothers remained in the same group or transitioned between groups over time. Predictors of group membership and transition will be also investigated (e.g., child disability status, child behavior problems).

**Discussion:** Using a measure of depressive symptoms with a non-clinical sample of mothers of children with or without ID, four groups of mothers were identified, and these groups were present across the 9 time points measured. Analyzing the stability/transition of mothers within/between groups, and possible predictors for group membership, has the potential to extend the current literature base by determining whether different types of mothers are differentially affected by their children with or without ID.

**References/Citations:**

Paper Title: Parent Assisted Social Skills Treatment in Early Childhood: The UCLA PEERS® For Preschoolers Program

Authors: Elizabeth Laugeson6, Mi Na Park6, Yasamine Bolourian1, Jennifer Sanderson7

Introduction: Social impairments characteristic of Autism Spectrum Disorder (ASD) are evident in early childhood and are likely to worsen with development (Rao, Beidel, Murray, 2008). Despite the growing popularity of parent training and early intervention programs, few evidence-based interventions exist that explicitly address the development of social skills in early childhood and actively integrate parents into treatment (DeRosier, Swick, Davis, McMillen, & Matthews, 2011).

Objective: The purpose of this study was to assess the feasibility and efficacy of a parent-assisted social skills intervention for preschool children with ASD using a randomized controlled design.

Methods: Children between four to six years of age diagnosed with ASD without intellectual disabilities participated in this study with their parents. Treatment involved the adaptation of an existing empirically supported social skills intervention for young children with ASD (Sanderson & Laugeson, 2008) by integrating treatment elements of the UCLA Program for the Education and Enrichment of Relational Skills (PEERS®; Laugeson & Frankel, 2010), an evidence-based social skills treatment for youth with ASD.

Families attended weekly 90-minute sessions delivered for 16 weeks in a small group format. Child sessions consisted of puppet-facilitated didactic lessons targeting key social skills, role-playing demonstrations, and play-based behavioral rehearsal of skills related to making and keeping friends. Parent sessions consisted of parent education and training, including review of socialization homework assignments, didactic lessons, and in-vivo performance feedback from the treatment team while social coaching their children during play-based activities.

Blinded behavioral observation of autism symptoms and social functioning were assessed at pre- and post-intervention using the Autism Diagnostic Observation Schedule - Second Edition (ADOS-2; Lord et al., 2012). Other treatment outcome measures completed by parents and independent blinded teachers at pre- and post-treatment included the Social Responsiveness Scale, Second Edition (SRS-2; Constantino, 2012), the Social Skills Improvement System (SSIS; Gresham & Elliot, 2008), and the Quality of Play Questionnaire (QPQ; Frankel & Mintz, 2010).

Results: Difference scores (DS) were calculated to examine changes in social functioning following treatment. Preliminary findings from paired sample t-tests of the open trial reveal a significant decrease in autism symptoms on the ADOS-2 from a moderate range of autism symptoms to a low range following treatment (p<.05), as measured by blinded clinical raters. Additionally, significant improvements in overall social responsiveness on SRS T-scores were observed in the areas of increased social communication (DS=5.1; p<.05), social awareness (DS=6.2; p<.05), social motivation (DS=6.5; p<.05), and decreased repetitive behaviors / restricted interests (DS=7.3; p<.05). Improvements in overall standardized scores of social skills (DS=4.9; p<.05) and decreased problem behaviors (DS=4.7; p<.05) on the SSIS were also observed, along with increased frequency of play dates on the QPQ (p<.05). No other statistically significant changes were observed across the outcome measures.

Discussion: Findings from the current study address a gap in the research literature by demonstrating the benefit of a parent-assisted social skills intervention in early childhood for youth with ASD.

6 UCLA Semel Institute for Neuroscience and Human Behavior
7 Florida Atlantic University
References/Citations:


**Symposium Title:** Parent-Implemented Spoken Language Intervention for Boys with FXS: A Naturalistic Language Intervention Delivered by Distance Teleconferencing

**Chair:** Andrea McDuffie

**Discussant:** Nancy Brady

**Overview:** There are few published studies of behavioral interventions designed to address the spoken language challenges experienced by boys with fragile X syndrome (FXS), the leading inherited cause of intellectual disability. In addition to significant cognitive delays, boys with FXS display phenotypic characteristics that include inattention, perseveration, tangential speech, social anxiety, and escape-maintained challenging behaviors. All of these behaviors are likely to interfere with the types of sustained interactions with conversational partners that are needed to support language learning. We recently examined the promise of a narrative-based, parent-implemented language intervention designed for adolescent boys with FXS (McDuffie, Bullard, Machalicek, Nelson, Mello, Tempero-Feigles, Castignetti & Abbeduto, in press). Using a multiple baseline design across three parent/child dyads, the goal of the intervention was to teach mothers to use three naturalistic language support strategies when interacting with their sons during shared story-telling. In addition to training mothers to act as their child’s interventionist, the intervention was unique in that intervention sessions were delivered into the home by means of distance teleconferencing. The intervention had a positive impact in that mothers increased their use of targeted intervention strategies and all three boys increased the number of different vocabulary words used during generalization sessions at the post-treatment. Two out of three boys also showed increases in syntactic development (MLU) as the result of the intervention.

**Paper 1 of 4**

**Paper Title:** Parent-Implemented Spoken Language Intervention for Boys with FXS: Feasibility of Assessment via Distance Teleconferencing

**Authors:** Lauren Bullard, Amanda Kwiatkowski, Melissa Mello, Andrea McDuffie, Leonard Abbeduto

**Introduction:** In addition to significant cognitive delays, boys with FXS display phenotypic characteristics that include inattention, perseveration, tangential speech, social anxiety, and escape-maintained challenging behaviors. Despite these challenges, families affected by such low incidence disorders often have limited access to center-based intervention programs.

We examined the effects of a spoken language intervention for three boys with FXS and their mothers. The intervention, including pre- and post-treatment assessment sessions, was delivered entirely in the family home via distance video-teleconferencing so that families were not restricted by geographical location to services. The current presentation is focused on the feasibility of collecting pre-and post-assessment data by means of distance teleconferencing. We addressed the following research questions:

1. Are pre/post observational measures collected via distance sensitive to changes in child spoken language and behavior after delivery of a parent-implemented intervention?
2. Are pre/post observation measures collected via distance sensitive to changes in maternal well-being?

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3. Are distance-collected observational measures more sensitive to change in comparison to parent-report questionnaires in measuring change in child behavior and maternal well-being?

**Methods:** Families were mailed a MacBook laptop computer, a standardized set of toys, and questionnaires. Mother’s completed the Vineland Adaptive Behavior Scales-II as a measure of child’s adaptive behavior. Dyads were observed via distance teleconferencing interacting in three contexts: (1) A semi-structured play sample using a standard set of developmentally appropriate toys; (2) Mothers were asked to make a snack with their child; (3) Mothers were asked to participate with their child in the telling of a wordless picture book. These samples were coded for parent use of targeted language support strategies and child nonverbal and verbal communication acts. Child symptoms of autism were also assessed from these sampling contexts using the Childhood Autism Rating Scale (CARS). In addition, mothers completed the Social Communication Questionnaire (SCQ) and the Aberrant Behavior Checklist (ABC). These measures along with the parent-child interaction were completed both pre- and post-treatment and will be used to assess changes in parent language, child language, and child behavior. Information also was gathered to assess maternal psychological well-being using the Symptom Checklist-90-R (SCL-90-R), the Parenting Sense of Competence Scale (PSOC), and the Parenting Stress Index (PSI). Lastly, mothers participated in the collection of the Five Minute Speech Sample (FMSS), which yields a measure of the emotional “climate” experienced by the child. These questionnaires and the FMSS sample will also be collected at the post-treatment as a measure of change in maternal well-being.

**Results:** Participants included three boys with FXS age 5- to 7-years old and their mothers who were participating in a parent-implemented language intervention. Child participants had a mean adaptive behavior composite score of 76 (range: 71-82). Preliminary results from Dyad 1’s parent-child interaction show promising evidence that both child language and behavior improved from pre- to post-treatment and that mothers were able to generalize language strategies to contexts beyond shared story telling. Information collected via the parent-child interaction will be compared to questionnaires regarding child behavior. Additionally, results from maternal questionnaires and the FMSS will be reported to assess changes in maternal well-being and the mother-child relationship.

**Discussion:** Given the small sample size, results reported in this section are best viewed as a test of feasibility in collecting information regarding pre- and post-treatment outcomes for individuals. While data collection is still underway, preliminary results indicate an improvement in child spoken language and behavior in parent-child observations collected via distance. These preliminary findings show that not only do the language strategies targeted during the intervention generalize to contexts such as play, but also show promising support for assessments collected via distance.

**Paper 2 of 4**

**Paper Title:** Parent-Implemented Spoken Language Intervention for Boys with FXS: The Role of Positive Behavior Supports

**Authors:** Melissa Mello¹, Lauren Bullard¹ ³, Amanda Kwiatkowski¹, Andrea McDuffie¹, Leonard Abbeduto¹

**Introduction:** There are few published studies of behavioral intervention designed to address challenges faced by boys with fragile X syndrome, the leading inherited cause of intellectual disability. In addition to significant cognitive delays, boys with FXS display phenotypic characteristics that include inattention, perseveration, tangential speech, social anxiety, and escape-maintained challenging behaviors. All of these behaviors are likely to interfere with the types of sustained interactions with conversational partners that are known to support language learning. These behaviors are also likely to interfere with the successful implementation of language intervention activities that require child engagement and active participation in order to be effective.
2016 Gatlinburg Conference Symposium

Applied behavior analysis (ABA) involves the systematic use of a variety of principles and techniques to bring about positive and meaningful change to behavior. Although the use of behavior analytic principles is well supported by research and widely applied to people with developmental delays, such as ASD, ABA has been less widely applied to children with FXS. The current study reports the results of a parent-implemented language intervention that targeted the spoken language skills of three young boys with FXS. Within the context of a naturalistic parent-implemented language intervention, a Board Certified Behavior Analyst taught mothers to use behavior analytic intervention strategies to decrease child challenging behaviors which interfered with the mothers’ attempts to engage her child in intervention activities. Mothers received education sessions and supported coaching from the BCBA who worked in collaboration with the team of speech/language pathologists. We addressed the following research questions:

1. Relative to baseline sessions, did mothers increase their use of recommended behavior interventions?
2. Relative to baseline sessions, did children decrease their frequencies of challenging behaviors to gain parent attention or escape activity?
3. Relative to baseline sessions, do children increase the level of engagement in the book reading task over time?

Methods: Three boys with a confirmed diagnosis of FXS and their biological mothers participated in the parent-implemented language intervention. Prior to beginning intervention to target language strategies, mothers received a parent education session focused solely on implementing behavior strategies (e.g., token economy, positive reinforcement). Mothers then completed a practice session with their child where they were coached on the behavior strategies. Throughout the language intervention, mothers were coached on behavior intervention strategies during the session and were provided with follow up strategies live after the call and/or via e-mail follow up.

Results: Preliminary results indicated that, relative to baseline, mothers increased their use of the behavior intervention strategies and children decreased their use of challenging behaviors and increased their level of engagement. Additionally, as demands increased (such as length of session or expected engagement from child) child challenging behavior decreased and/or was successfully managed by parent to maintain participation in the session. Data collection and coding are still underway.

Discussion: Boys with FXS are often overlooked when it comes to the use of proven applied behavior analysis techniques. They struggle with maladaptive behaviors that serve to get them attention or out of a low preferred situation. It can also be difficult for them to attend to tasks, especially low preferred tasks, for increased amounts of time. As the result of this intervention, mothers learned a set of strategies they could use to decrease the amount of challenging behaviors their children were displaying, primarily ones that were maintained by mothers attention or escape. Boys increased the length of time they were able to engage in the book reading activity and expressed more positive social behavior with their mothers over time. Clinical implications and future directions will be discussed.

Paper 3 of 4

Paper Title: Parent-Implemented Spoken Language Intervention for Boys with FXS: Effects on Maternal Strategy Use

Authors: Andrea McDuffie1, Lauren Bullard1, 3, Amanda Kwiatkowski1, Melissa Mellow1, Leonard Abbeduto1

Introduction: There are few published studies of behavioral intervention designed to address the spoken language challenges faced by boys with fragile X syndrome, the leading inherited cause of intellectual disability. In addition to significant cognitive delays, boys with FXS display phenotypic characteristics that include inattention, perseveration, tangential speech, social anxiety,
and escape-maintained challenging behaviors. All of these behaviors are likely to interfere with the types of sustained interactions with conversational partners that are known to support language learning.

We examined the effectiveness of a parent-implemented spoken language intervention for three young boys with FXS and their mothers. The goal of the intervention was to teach each mother a set of strategies for interacting with her child in ways that would support spoken language development. Specifically, the intervention targeted the use of four language support strategies: (a) modeling story related vocabulary and grammar, and (b) use of open-ended questions, (c) expansions, and (c) intonation prompts. We addressed the following research questions:

1. Relative to baseline, did mothers increase their use of language strategies; including expansions, open ended questions, and intonation prompts?

2. Relative to baseline, did mothers increase their overall story related talking?

**Methods:** The intervention used a single subject multiple baseline design across the three parent/child dyads. An SLP and BCBA provided real time coaching to the mother as she interacted with her child during intervention sessions. Each mother selected 12 wordless 15-page picture books which were digitized and loaded onto an iPad. The language intervention consisted of 2 didactic parent education sessions to introduce language strategies, followed by 12 weekly parent/child coaching sessions to support mothers in building their capacity to provide the intervention to their children. Additionally, mothers independently completed one homework session each week and received feedback from the SLP using video clips made from the homework sessions. Finally, a data collection session was completed weekly. All sessions were implemented via distance teleconferencing (e.g. Skype). Homework and data collection sessions were transcribed using Systematic Analysis of Language Transcripts (SALT). Transcripts were coded for maternal use of language strategies such as use of open ended questions, expansions, and intonation prompts. A composite score of language strategies and parent models of story related vocabulary and grammar was calculated to represent overall story related talking.

**Results:** Preliminary data from Dyad 1 indicate that the mother increased her overall use of language strategies per book from baseline to intervention. Specifically, her use of open-ended questions increased from baseline (M=1) to intervention (M=19); expansions increased from baseline (M=5) to intervention (M=45); and intonation prompts increased from baseline (M=1) to intervention (M=17). The mother in Dyad 1 also increased her overall story related talking per book from baseline (M=7) to intervention (M=141). Data collection and coding are still underway.

**Discussion:** Social interactionist theories of language development support the premise that language is learned best when children can interact with more competent communicative partners. In addition, the quality and quantity of verbal language input that children receive within the context of communicative interactions is critical for supporting language growth. Providing rich verbal language input to their children is a challenge for mothers of boys with FXS as these children, in addition to language delays, have other challenging behaviors that interfere with their ability to sustain the kinds of frequent back and forth interactions that support language growth. As the result of this intervention, mothers learned a set of strategies they could use to sustain a conversational interaction with their child within the context of shared book reading, a common interactive context for typically developing children. Clinical implications will be discussed.
**Paper Title:** Parent Implemented Spoken Language Intervention for Boys with FXS: Effects on Child Spoken Language

**Authors:** Amanda Kwiatkowski¹, Lauren Bullard¹, ³, Melissa Mello¹, Andrea McDuffie¹, Leonard Abbeduto¹

**Introduction:** There are few published studies of behavioral intervention designed to address the spoken language challenges faced by boys with fragile X syndrome, the leading inherited cause of intellectual disability. In addition to significant cognitive delays, boys with FXS display phenotypic characteristics that include inattention, perseveration, tangential speech, social anxiety, and escape-maintained challenging behaviors. All of these behaviors are likely to interfere with the types of sustained interactions with conversational partners that are known to support language learning.

We examined the efficacy of a parent-implemented spoken language intervention for three young boys with FXS and their mothers. The goal of the intervention was to teach each mother a set of strategies for interacting with her child in ways that would support their child's spoken language development. Child outcome measures included mean length of utterance, number of different vocabulary words, and number of story related utterances used during intervention and generalization sessions. We addressed the following research questions:

1. As the result of participation in a parent implemented language intervention, do children increase their vocabulary diversity and grammatical complexity?

2. Relative to baseline, do children increase their overall story related talking during a telling of a wordless picture book?

**Methods:** During the intervention, dyads participated in a weekly coaching session during which an SLP and BCBA provided in vivo support and guidance as the mother interacted in real time with her child. Additionally, mothers independently completed one homework session each week and received feedback from the SLP using video clips made from the homework sessions. Finally, a data collection session was completed weekly. A different wordless picture book was used during each week of the intervention. Homework and data collection sessions were transcribed using Systematic Analysis of Language Transcripts (SALT). Transcripts were coded for child outcome measures including: mean length of utterance (MLU), number of different words (NDW), and number of child's complete story related utterances.

**Results:** Preliminary results for the child participant in Dyad 1 show an increase in mean length of utterance (MLU) from 1.6 words to 2.5 words from baseline to intervention. Mean number of different words (NDW) also increased from 17 words per book in baseline to 95 different words per book in intervention. Lastly, the child in Dyad 1 had a mean of 14 story related utterances during baseline sessions. The same child had a mean of 126 story related utterances during intervention sessions.

**Discussion:** Shared story telling provides a naturalistic context within which mothers of boys with FXS can support their children's language development both during and following a parent implemented language intervention. In this pilot study mothers used four different strategies to provide their children with models of more diverse vocabulary and developmentally advanced syntax. Additionally, mothers used behavioral support strategies to increase child engagement and active participation in intervention activities. Preliminary data suggests that, relative to baseline sessions, child participants increased their use of different vocabulary words and used more complex grammar both during intervention sessions and during generalization sessions. Future studies should examine whether this same approach can be used to support spoken language development in other groups of children with developmental delays.
Symposium Title: Self Injurious Behavior, Proto-Injurious Behavior, and Restricted and Repetitive Behavior in IDD and ASD: Contingencies, Precursors, and Developmental Context

Chair: Kristen Medeiros

Overview: This symposium will discuss various behavior problems from three perspectives: contingencies, precursors, and developmental contexts. The studies utilize a variety of samples and methods to answer questions about behavior problems in order to assist in developing successful interventions. These findings are useful for clinical researchers, educators, or policy makers in the field of intellectual and developmental disabilities and Autism.

Paper 1 of 3

Paper Title: "Contagious" Self-Injurious Behavior among Individuals with Intellectual And Developmental Disabilities in Community Settings

Authors: Andrea B. Courtemanche, Blair P. Lloyd, Johanna L. Staubitz, Sherry Crossley

Introduction: Self-injurious behavior (SIB) can be a chronic condition among individuals with intellectual and developmental disabilities (IDD). SIB may be a learned behavior maintained by environmental consequences (e.g., staff attention) (Carr, 1977). Using sequential analysis methodology, several studies have documented a sequential dependence between instances of SIB (i.e., contagious SIB) rather than dependence between SIB and social consequences (e.g., Marion, Touchette, & Sandman, 2003). That is, rather than SIB resulting in a consistent social consequence, one instance of SIB is likely to be followed by another instance of SIB. Sequentially-dependent SIB has also been associated with altered patterns of endorphins and other hormones, suggesting that SIB may be maintained by biological factors rather than environmental (Sandman et al., 2008). Because many of the participants in these studies lived in institution-like settings, it is unclear whether the absence of social contingencies may have been due to relatively low rates of social attention in that environment. The purpose of the present study was to use sequential analysis methodology to assess behavior-behavior (SIB-SIB) and behavior-environment (SIB-staff attention) contingencies for a group of individuals with IDD living in community settings.

Methods: Seven individuals (ages 15-50) withIDD and chronic SIB were recruited to participate. Individuals were videotaped during their daily routines at home and in the community. A continuous, timed-event recording system was used to code videos for the frequency of SIB and the frequency and duration of staff attention and participant engagement in functional activities. Participant and staff behavior were analyzed for frequency, duration, inter-observer agreement, and sequential dependencies (i.e., Yule’s Q). An event lag with contiguous pause approach (Lloyd, Yoder, Tapp, & Staubitz, in press) was used to analyze sequential dependencies.

Results: For many participants, a sequential pattern of SIB was identified. Thus, for some of these individuals, SIB was more likely to be followed by another instance of SIB than by staff attention. The best predictor of SIB was a previous instance of SIB. Additionally, sequential patterns varied based on idiosyncratic topographies of SIB.

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2 University of Saint Joseph
3 Vanderbilt University
**Discussion:** The results of the current study suggest that some participants’ SIB may be maintained by non-social consequences. Additionally, these results highlight the possibility that multiple topographies of SIB may serve multiple functions. Future research should evaluate the relationship between sequential associations of SIB in natural contexts, behavioral function as determined by a functional analysis and potentially relevant biological factors to better understand the causal mechanisms of chronic SIB.

**References/Citations:**

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**Paper 2 of 3**

**Paper Title:** Stereotyped Behaviours as Precursors of Self-Injurious Behaviours: A Longitudinal Study with Infants and Toddlers at Risk for Developmental Delay

**Authors:** Johannes Rojahn⁴, Lucy Barnard-Brak⁵, Kristen Medeiros¹, Stephen Schroeder⁶

**Introduction:** The notion that stereotypic behaviours may be functional (Guess & Carr, 1991) and structural (Berkson, Tupa, & Sherman, 2001) precursors of self-injurious behaviour (SIB) has been considered in the past, but the available empirical evidence is still inconsistent and ambiguous.

**Methods:** In a longitudinal study, we collected data on stereotypic behaviour and SIB from 160 infants and toddlers who were at-risk for developmental delay. Interviews were conducted with parents at three time points during a one-year span using the Behaviour Problems Inventory-01 (BPI-01; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001), which contains subscales for SIB and stereotyped behaviour. We used growth modeling to estimate linear trends in several models. Model fit was evaluated according to a combination of fit statistics as is recommended in structural equation or latent variable modeling approaches such as latent growth modeling.

**Results:** In examining the relationship between stereotyped behaviours and SIB across time, the model that represented earlier stereotyped behaviour as predicting later SIB fit the data better than the other models.

**Discussion:** The findings corroborate the notion that stereotyped behaviours can be a precursor of SIB. If replicated by other studies, it makes a case for considering early intervening with stereotyped behaviour as a SIB prevention strategy.

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⁴ George Mason University  
⁵ Texas Tech University  
⁶ University of Kansas
References/Citations:


Paper 3 of 3

Paper Title: Developmental Trajectories of SIB and RRB in Children with Autism

Authors: Cristan Farmer#, Lisa Joseph#, Audrey Thurm#

Introduction: Differential expression of subtypes of restricted and repetitive behavior (RRB) in autism spectrum disorder (ASD) across childhood has been documented in both cross-sectional and longitudinal data (Esbensen, Seltzer, Lam, & Bodfish, 2009; Richler, Huerta, Bishop, & Lord, 2010). Very little is known about the childhood developmental course of self-injurious behavior (SIB) in ASD, although cross-sectional data suggest that the behaviors are more common in younger children (Baghdadli, Pascal, Grisi, & Aussilloux, 2003). The elucidation of the developmental trajectories of RRB and SIB, and their relationship over time, may help to identify subgroups of children with ASD with shared etiology.

Methods: The Repetitive Behavior Scale-Revised (RBS-R; Bodfish, Symons, Parker, & Lewis, 2000; using the Lam & Aman, 2007 structure) was completed up to six times by parents of 106 children aged 2-9 years enrolled in a longitudinal naturalistic study of DSM-IV-TR autistic disorder. Latent class growth curve modeling was used to characterize developmental trajectories of scores on the RBS-R, including Stereotyped Behavior, Restricted Interests, Self-Injurious Behavior, Compulsive Behavior, and Ritualistic/Sameness Behavior. Finally, we plan to estimate joint trajectory models to characterize the developmental relationships among the constructs.

Results: Baseline latent growth curves were fit to the data from each subscale of the RBS-R. Subscales representing lower-order behaviors (i.e., SIB, Stereotyped Behavior, and Restricted Behavior) were best represented by intercept-only models, while the higher-order behavior subscales (i.e., Compulsive Behavior and Ritualistic/Sameness Behaviors) were best represented by quadratic growth curves. However, chi-square tests of model fit were significant for all models, indicating lack of fit. We proceeded with latent class mixture model analysis, using the best-fit latent growth curve model as the baseline. Requesting up to four classes and progressively releasing model constraints, we selected the best-fitting model using several standard measures of relative and absolute fit. For all subscales, the selected latent class mixture model was significantly better fit to the data than the latent growth curve. Two-class models best fit the lower-order behaviors: SIB: Low/steady, 94%; low/rising, 6%; Stereotyped: Low/steady, 73%; low/increasing, 27%; Restricted Interests: low/rising, 81%; high/decreasing, 19%. The remaining subscales were best fit by three-class models: Compulsive (low/decreasing, 67%; moderate/increasing, 21%; high/decreasing, 12%) and Ritualistic/Sameness (low/steady, 84%; low/rising, 10%; moderate/increasing, 6%).

# Pediatrics & Developmental Neuroscience Branch, National Institute of Mental Health, National Institutes of Health
Discussion: Non-mixture latent growth curves did not sufficiently describe the development of RBS-R scores between the ages of 2 and 7 years in this sample of children with ASD. Models with two or three classes of varying trajectories better described the data. We are currently fitting joint-trajectory models, to explore whether the profile of development in RRB is a useful phenotypic indicator. Finally, we will explore predictors of class membership, such as medical information (e.g., genetic conditions, prenatal/perinatal complications, other health problems) and behavioral data (e.g., severity of non-RRB ASD symptoms, cognitive level, adaptive functioning). All of these results, as well as general methodological issues, will be the focus of discussion.

References/Citations:

 Symposium Title: Life Course Predictors of Social Functioning in Individuals with Autism Spectrum Disorders

Chair: Marsha Mailick

Overview: Human beings are inherently social. From expressing oneself through various gestures, facial expressions, and other nonverbal movements to being able to represent other peoples' minds, our ability to socially connect and interact is an important component of our lives and our well-being. Impairments of social behaviors can not only have severe repercussions on how an individual takes part in everyday social interactions, but, more importantly, impact their quality of life. Such deficits and social 'dysfunction' are hallmark characteristics of autism spectrum disorder (ASD). Complex and heterogeneous in nature, individuals diagnosed with this lifelong developmental disability typically have difficulty in several domains of social functioning, while recent research has additionally suggested that these deficiencies may have a greater impact over time. Despite the known impact of impairments in social functioning on the lives of individuals with ASD, little is known about how social functioning develops across the life course in ASD and how it is altered by biological and behavioral influences. To study this critical knowledge gap, the abstracts of this symposium utilized data from several ongoing studies of ASD to specifically explore the stability of measures of social functioning across a wide age range of individuals. This symposium also explores how factors of parental social functioning, levels of stress, language ability and the underlying neurobiology impact social functioning in ASD. Importantly, the findings from this symposium begin to elucidate critical relationships of observed social behaviors in ASD and provide preliminary insight to markers that may be beneficial in future intervention studies.

Paper 1 of 5

Paper Title: Validity Tests of the Social Responsiveness Scale (SRS) for Adults with Autism Spectrum Disorders: A Latent Factor Approach

Authors: Wai Chan, Leann Smith, Jinkuk Hong, Jan Greenberg, Marsha Mailick

Introduction: The Social Responsiveness Scale (SRS; Constantino & Gruber, 2005) was primarily developed to quantify severity of social impairment among children and adolescents affected by autism spectrum disorders (ASD) or other developmental disabilities. SRS has also been applied to recent investigations with adult samples, suggesting that it yielded good sensitivity (e.g., Bölte, 2012). Nevertheless, very few studies have examined the measurement properties of SRS specific to adults with ASD. SRS addressed five dimensions of social impairment, i.e., social awareness, social motivation, social communication, social cognition, and autistic mannerism. However, a total sum score approach is commonly used which collapsed SRS dimensions into an index score with higher score representing greater impairment. This validation study hence introduced a latent factor approach for better representing the underlying structure of SRS and sought empirical support for its psychometric properties in an adult sample with ASD. Both concurrent and predictive validity of the latent factor were examined.

Methods: The current sample used a subsample of a larger longitudinal study (Adolescents and Adults with Autism, AAA; Seltzer, Greenberg, Taylor, Smith, Orsmond, Esbensen, & Hong, 2011). Mothers of adult children with ASD (N = 237; M = 29.47, ranged from 18 - 57 years old) provided responses to SRS, and multiple ASD-related measures (i.e., Autism Diagnostic Interview-Revised, ADI-R; Scale of Independent Behavior-Revised, SIB-R; Vineland Screener, VS) at two time points with approximate 18 months apart. A single latent factor with all five SRS subscale scores loaded as indicators were modelled using confirmatory factor analysis (CFA) techniques. Model fit indices were examined. Its strong correlations in the expected direction with other ASD-related measures indicated high validity of the latent factor approach.

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2 Waisman Center, University of Wisconsin-Madison
Results: CFA results indicated that the latent factor of SRS provided good model fit to the after allowing residual covariances between social communication and motivation ($\chi^2(4) = 3.43, p = .49; \text{CFI} = 1.00; \text{RMSEA} = .00, 95\% \text{CI} [.00 .09]$). It was also highly correlated with ASD-related measures concurrently and prospectively (ADI-R: $r_s = .63$; SIB-R: $r_s \geq .55$, VS: $rs \geq -.57$). All correlation values were significant at $p < .001$.

Discussion: The latent factor of SRS demonstrated satisfactory model fit to the data, consistent with the multi-dimensionality of ASD-related instrument. The residual covariances were justified due to the conceptual overlap between social communication and motivation (Chevalier et al., 2012). The latent factor also demonstrated high validity, establishing psychometric properties of SRS specific to adult population with ASD.

Another strength of the latent factor approach is to account for measurement error of all SRS subscales, a statistical technique that ensures reliable estimates and reduction of false positive related to ASD (Heavner et al., 2014). Thus, the use of the latent factor approach was encouraged especially when adults with ASD are the study sample.

References/Citations:

Paper 2 of 5

Paper Title: Language and the Social Responsiveness Scale (SRS) in School-Age Children with High Functioning Autism

Authors: Philip Lai², Judy Reilly³

Introduction: Social behavior affects all aspects of one's life. Lacking the ability to process and effectively convey social information can lead to deficits in maintaining peer and family relationships, in addition to academic failure in school. In this study, the Social Responsiveness Scale (SRS), a parental questionnaire was used to investigate the relationship between language use and levels of social functioning in individuals with High Functioning Autism (HFA).

Methods: Participants in this study included 23 children with HFA between the ages of 7-14 years as well as their parents/guardian. In this investigation, a semi-structured naturalistic biographical interview was conducted as children sat at a table across from the interviewer. The subjects were asked a set of open-ended questions from a script on a range of everyday topics (e.g., "what did you do over the weekend?") and the interviewer subsequently followed up with questions and comments specific to the child's responses. The number of complex sentences was counted to determine the frequency of complex syntax. Complex sentences are multiple propositions that fall within a sentence intonation contour. Categories included: all coordinate sentences, verb complements, relative clauses, passive sentences, and adverbial clauses. To control for length, Rate of Complex

² San Diego State University
Syntax was calculated by taking the total number of complex constructions and dividing by the total number of child produced propositions in the conversation. A proposition was defined as a verb and its arguments; from a semantic perspective a proposition roughly corresponds to a single event. Social evaluations were also investigated. Social evaluative devices are used to attract and maintain the listener's attention through the narrator's attitude or perspective. Examples can include the use of emotional states and verbs, emphatics, intensifiers, character speech, direct quotes, asking questions, and sound effects.

Guardians completed the SRS, a 65-item inventory used to assess their children's social and communicative abilities. Guardians were instructed to select the rating or response that best fit their own interactions with or observations of the child.

Results: For measures of syntactic complexity, social awareness, social motivation, social cognition, and autistic mannerism did not correlate with the production of complex sentence constructions. A trend was observed for social communication, where fewer complex sentences were observed in children with greater social communication deficits, $r = 0.368$, $n = 23$, $p = 0.084$ (2-tailed). For the production of social evaluative devices, social awareness, social motivation, social communication, and social cognition did not correlate with this linguistic measure. Autistic mannerism did correlate, $r = 0.541$, $n = 23$, $p = 0.008$ (2-tailed), as more social evaluative devices were observed for children with higher scores on autistic mannerism.

Discussion: Our findings with respect to the production of complex sentences highlight communication deficits are still observed in children with HFA at school-age. Our result of higher uses of social evaluative devices in children with greater deficit with respect to autistic mannerism is more perplexing. This result may be due to our task, where the experimenter would ask a question and follow-up with additional questions based on the answers the child has given. The experimenter never cut the child off at any point in the conversation, allowing the child to speak on a particular topic which can last for many minutes. Due to this, a child can speak on a topic that interest them, perhaps using language that may be more engaging, resulting in greater uses of social evaluative devices. Future studies should examine communicative behaviors when children with HFA initiates a conversation compared to when they respond to their interlocutor's question.

Paper 3 of 5

Paper Title: Associations between Father and Mother Broader Autism Phenotype, Family Environment, and Adult Child's Autism Symptoms

Authors: Daniel Laxman², Leann Smith², Jan Greenberg¹, Marsha Malick¹

Introduction: Individuals with an autism spectrum disorder (ASD) experience impaired social functioning, communication difficulties, repetitive behaviors, and restricted interests. Studies have found that some parents of individuals with ASD exhibit subclinical levels of autistic traits—known as the broader autism phenotype (BAP)—in the form of social aloofness, pragmatic language, and rigidity (e.g., Hurley et al., 2007; Piven, 2001). Researchers are beginning to explore the association between parent’s BAP and the severity of their children’s autistic symptoms, but results have been mixed (e.g., Hasegawa et al., 2014; Maxwell et al., 2013; Sasson et al., 2013; Seidman et al., 2012). Past research suggests that genetic inheritance is a mechanism for the intergenerational transmission of autistic symptoms (Lyal et al., 2014), although the genetics of autism remain poorly understood and likely involve the interaction of hundreds of genes (Betancur, 2011). However, there is a great deal of evidence that the family environment has an effect on the severity of children's autism symptoms (e.g., Smith et al., 2014), and parental BAP may contribute to the family environment (e.g., Orsmond & Seltzer, 2009). The present study builds on previous research by exploring the association between parents' BAP characteristics, family environment, and the level of social impairment of adult children with ASD. Furthermore, analyses explored whether the association between parents' BAP and adult children's social impairment varied by the sex of the adult child.
**Methods:** The sample for this study was drawn from a larger study on adolescents and adults with ASD (Seltzer et al., 2011). Because autistic characteristics are thought to be transmitted in part through genetic inheritance, the sample was restricted to individuals with ASD (N=102; Age: M=27.37, SD=6.58, range=18-54) and their cohabitating biological parents. Hierarchical multiple regression analyses tested whether mother’s and father’s scores on the Broader Autism Phenotype Questionnaire (Hurley et al., 2007) and characteristics of the family environment (parents’ mutual support, mother’s depressive symptoms, and mother’s criticism) predicted adult children’s severity of social impairment (Social Responsiveness Scale; SRS; Constantino & Gruber, 2005). Analyses controlled for demographic and other characteristics (intellectual disability status, prior severity of autism symptoms) of the adults with ASD. Moderation analyses explored whether parents’ BAP interacted with the sex of the child in predicting the adult child’s social impairment.

**Results:** Results indicate that higher levels of fathers’ BAP characteristics were associated with greater social impairment for adults with ASD (standardized b=0.21, p=.017). The same association existed for mothers, but only for families of sons with ASD (b=0.27, p=.007). Of the family environment variables, parents’ mutual support was associated with lower levels of their adult child’s social impairment (b=–0.18, p=.026) while mother’s level of criticism was associated with higher levels (b=0.28, p=.001).

**Discussion:** These results confirm previous findings that father’s and mother’s BAP levels are associated with the degree of social impairment found in their children with ASD. Interestingly, for mothers, this association only held for families of sons with ASD. This finding could suggest that mother’s BAP characteristics reflect a greater genetic liability to autism and symptom severity for sons than for daughters. Alternatively, this finding could indicate that mother’s BAP characteristics may negatively affect the quality of the family environment, which in turn may influence son’s social impairment more than daughter’s. Additional analyses will explore these possibilities. Finally, characteristics of the family environment were significantly associated with children’s social impairment even after accounting for parents' BAP. This finding highlights the need to consider both characteristics of the family environment and parents' BAP in research and practice targeting social impairment in individuals with ASD.

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**Paper Title:** Correlates of Social Functioning in Adults with Autism Spectrum Disorder: The Role of Biological Stress Response and Perceived Stress

**Authors:** Lauren Bishop-Fitzpatrick, Carla Mazefsky, Nancy Minshew, Shaun Eack

**Introduction:** Poor social functioning is a central feature of autism spectrum disorder (ASD) and has far-reaching effects on multiple domains of life. Yet, we know little about modifiable factors that predict social functioning in this population. Preliminary work suggests that there may be differences in the underlying mechanisms that drive stress management in individuals with ASD and that these differences may be associated with adult outcomes. This study aimed to examine the biological and behavioral underpinnings of adult outcomes in ASD by (1) identifying differences in stress response among adults with ASD and healthy volunteers; and (2) examining the relationship between stress response and social functioning adults with ASD. We hypothesized that adults with ASD would experience greater biological stress, perceived stress, and stressful life events than healthy volunteers and that there would be a significant relationship between stress and social functioning in adults with ASD.

**Methods:** Cross-sectional data were collected from 40 adults with ASD and 25 healthy volunteers. Participants with ASD were ages 18-44 (M=24.2), had intelligence quotient (IQ) scores from 80-132 (M=106.3), and 90.0% male. Healthy volunteers were

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ages 18-32 (M=25.1), had IQ scores from 82-138 (M=110.6), and 84.0% male. Repeated measures of systolic blood pressure (SBP), diastolic blood pressure (DBP), and heart rate (HR) were taken during a social stress challenge task, while salivary cortisol was collected before and after the task. Measures also assessed perceived stress (Perceived Stress Scale), stressful life events (Stress Survey Schedule), social disability (Social Adjustment Scale-II), daily living skills (Waisman Activities of Daily Living), and social impairment (Social Responsiveness Scale). Analyses examined group differences between adults with ASD and healthy volunteers on biological stress response variables (SBP, DBP, and HR growth coefficients and salivary cortisol change scores) and self-reported stress variables (perceived stress and stressful life events). The relationship between biological stress response and psychosocial stress variables and social functioning variables (global functioning, social impairment, social disability, and daily living skills) was analyzed using hierarchical multiple regression procedures.

Results: Results indicated that adults with ASD experienced significantly more perceived stress, $F(1, 63)=23.13, p<.001$, and stressful life events, $F(1, 63)=25.90, p<.001$, and greater SBP reactivity, $F(1,63)=4.95, p<.05$ but did not differ from healthy volunteers in terms of DBP reactivity, HR reactivity, or cortisol reactivity. Results of regression analyses indicated that perceived stress, $\beta=0.07, t(35)=3.16, p<.05$, and stressful life events, $\beta=0.02, t(35)=3.20, p<.05$, significantly predicted social disability, when controlling for age, IQ, and treatment exposure. However, none of the biological stress response variables significantly predicted any social functioning variables, and perceived stress and stressful life events did not predict social impairment or daily living skills.

Discussion: These results suggest that, while adults with ASD experience both greater systolic blood pressure reactivity and greater perceived stress and stressful life events than healthy volunteers, the perception of life as stressful predicts social disability in this population while measured biological response to stress does not. However, directionality is unclear. Future research should confirm directionality and examine interventions that might improve social functioning by helping adults with ASD experience stress differently.

**Paper 5 of 5**

**Paper Title:** Neurobiological Predictors of Social Function in Autism Spectrum Disorders

**Authors:** Douglas Dean$^2$, Brittany Travers$^2$, Brandon Zielinski$^6$, Molly Prigge$^6$, Erin Bigler$^7$, Nicholas Lange$^8$, Janet Lainhart$^2$, Andrew Alexander$^2$

**Introduction:** Impairments of social function and communication are defining characteristics of autism spectrum disorders (ASD) that make it difficult for individuals with ASD to engage in social interactions. As the brain is responsible for much of an individual's cognitive and behavioral processes, it has been hypothesized that specific brain regions responsible for social function may be altered in individuals with ASD. Specifically, the white matter microstructure is essential to the normative functioning, as it links discrete regions together and coordinates highly efficient, temporally precise communication between them. However, little is known about the role white matter has on social functioning in this population. This study therefore sought to examine relationships between measures of white matter microstructure and social function in ASD and identify regions that differed from age-matched controls. We hypothesized that individuals with ASD would have altered microstructure in neighboring white matter areas associated with social function, including occipital and temporal brain regions.

**Methods:** MRI Acquisition: Participants for this study consisted of 100 males with ASD and 56 age-matched males with typical development (TD) between 3 and 42 years of age. Longitudinal diffusion tensor imaging (DTI) data were acquired from each

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$^8$ Harvard Medical School
participant, totaling 434 observations (287 ASD, 147 TD). Images were corrected for distortion and head motion and maps of fractional anisotropy (FA), mean diffusivity (MD), and radial diffusivity (RD) were calculated, however, only FA was used herein. The Social Responsiveness Scale (SRS) was additionally collected and the total raw SRS score was used as a measure of overall social impairment. Analysis: Voxel-wise longitudinal mixed effects models were constructed and fit to the data to examine the relationships between the white matter microstructure (i.e. FA) and social function (i.e. SRS total raw score). Additional covariates of age, an index of total motion during the scan, and head coil, were included in the models. Correction for multiple comparisons was performed using the False Discovery Rate (FDR) and significance was defined as p<0.05, FDR corrected.

Results: Results from the voxel-wise longitudinal mixed effects modeling indicated the ASD group to have widespread significant (p<0.05, FDR corrected) alterations of FA. Specifically, reduced FA in white matter regions of the left hemisphere inferior longitudinal fasciculus, left inferior fronto-occipital fasciculus, bilateral corticospinal tract, and inferior regions of the internal capsules, were found to be predictive of higher SRS total raw measures.

Discussion: Our findings suggest that white matter regions implicated in brain regions associated with social function are altered in ASD individuals, compared to individuals with typical development. As these white matter pathways are involved in brain connectivity, this study suggests that altered brain connectivity may be a central feature to alterations of social functioning in ASD. Furthermore, this study raises new questions about the underlying factors of these white matter alterations. Are these alterations caused by abnormalities to neurons and synapses or are they a result of aberrant myelination? Future studies that utilize more sophisticated imaging strategies should examine such questions. Moreover, the specific brain regions implicated in this study could serve as targets for future interventions and preventative treatments in order to assess whether such interventions can modify the white matter microstructure and if such changes result in improved social functioning.
Symposium Title: Novel Outcome Measures for Intellectual Disability Clinical Trials

Chair: David Hess¹

Discussant: Leonard Abbeduto¹

Overview: Clinical trials of individuals with intellectual disabilities are challenging, in part due to the dearth of well-validated, reliable and sensitive outcome measures for the population. This symposium will present research on two cognitive batteries and a unique eyetracking protocol targeting social gaze. Many of these measures have already been embedded in fragile X targeted treatment trials, affording an opportunity to examine their sensitivity to change.

Paper 1 of 3

Paper Title: The NIH Toolbox Cognitive Battery for Intellectual Disabilities: Preliminary Feasibility Studies and Future Directions

Authors: David Hess¹, Stephanie Sansone¹, Elizabeth Berry-Kravis², Richard Gershon³, Karen Riley⁴, Andrea Schneider¹, Crystal Crestodina², Kelly Rhodes², Dena Oaklander²

Introduction: Although intellectual disabilities (ID) have been considered to be lifelong, with little promise for meaningful recovery of cognitive functions, recent advances in the understanding of the underlying genetic and neurobiological abnormalities of syndromic forms of several disorders such as fragile X (FXS) and Down syndromes (DS), as well as advances in targeted pharmacological and behavioral treatments, provide evidence that substantial gains are possible and may contribute to real life improvements in daily functioning. However, thus far, several well-powered controlled trials of drugs proven to be highly effective in animal studies of FXS have failed to show efficacy on primary clinical targets of maladaptive behavior. Although investigators are now focusing on cognition as a primary clinical target for future trials, there is little consensus on just how cognitive improvements should be objectively measured. Here we present our preliminary research efforts to establish and validate a cognitive battery for intellectual disabilities based on the NIH Toolbox Cognitive Battery (NIH-TCB; www.nihtoolbox.org), a web-based and touch-screen response system normed on children and adults 3-89 years, with a primary focus on its use as a series of cognitive endpoints for targeted pharmacological trials and other treatment studies.

Methods: We completed two feasibility-oriented pilot studies of NIH-TCB measures. The first focused on two executive function (EF) measures, Flanker and Dimensional Change Card Sort (DCCS) in 31 patients with FXS (20 at Davis, 11 at RUMC; 27 male) between the ages of 5 and 36 years (mean=19.3), a mean IQ of 47.0 and mean mental age of 5 years 3 months. The second study utilized all seven TCB measures (addressing EF, working memory, vocabulary, single letter/word reading, processing speed, and episodic memory) in 22 patients with FXS (16 male; mean age 20.5 years) and 28 with DS (15 male; mean age 16.3 years), with mental ages ranging from 2 to 10.

Results: In Study 1, on Flanker, participants with FXS made significantly more errors and demonstrated longer reaction times for incongruent than congruent trials (59% vs. 80% correct, p = 0.003; 2363 ms vs. 1889 ms, p = 0.002). On DCCS, they made more errors on trials involving a switch (change cognitive set) than without a switch (66.2% vs. 82.0% correct; p < 0.001), reflecting attention and inhibitory control effects of the tasks. Flanker and DCCS demonstrated substantial range and variability, without flooring or ceiling, and showed modest Spearman correlations with full scale IQ (Flanker accuracy, $r = 0.65$, $p = 0.003$; DCCS

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⁴ Denver University
switch, \( p = 0.52, p = 0.03 \) ), good to strong sensitivity to mental age (Flanker accuracy, \( p = 0.89, p < 0.001 \); DCCS Switch, \( p = 0.68, p = 0.001 \). In Study 2, 66% (working memory) to 92% (vocabulary and reading) of participants were able to provide valid data. Test-retest reliability (N = 17; Mean = 37 days; range 13 - 95) intra-class coefficients ranged from .65 (DCCS) to .99 (reading) with average of .81. Syndromic differences (covarying for age/mental age) were observed for processing speed (DS<FXS, \( p = .01 \)) and vocabulary (DS<FXS, \( p = .06 \)) only. The NIH-TCB composite Z score correlated well with available IQ scores (N = 24, \( r = .73, p < .001 \)) and less well with adaptive behavior scores (N=24, \( r = .35, p = .09 \)).

**Discussion:** The NIH-TCB shows promise as a battery of outcome measures for ID, however adaptations for lower functioning and younger participants are needed, and cross-measure validation, more controlled test-retest, and sensitivity-to-change studies are required. A large multisite, NICHD funded study supported by the data presented here is underway to accomplish these aims and will be discussed.

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**Paper 2 of 3**

**Paper Title:** The KiTAP as an Outcome Measure for Executive Function in Clinical Trials for Fragile X Syndrome

**Authors:** Elizabeth Berry-Kravis\(^5\), David Hess\(^1\), Jonathan Rubin\(^5\), Yaron Daniely\(^5\), Crystal Crestodina\(^2\), Andrew Knox\(^2\), Andrea Schneider\(^5\), Mary Jacina Leigh\(^1\), Kyoungmi Kim\(^3\), Randi Hagerman\(^1\)

**Introduction:** Although individuals with fragile X syndrome (FXS) have prominent executive function and attention deficits, it has been challenging to find measures of executive function or attention that can be done successfully and produce valid scores across the range of functional ability observed in FXS. The Test of Attentional Performance for Children (KiTAP) is an 8 subtest computer-based pictorial battery of executive function tests with an enchanted-castle theme. In a pilot study, the KiTAP was administered to 36 subjects with FXS across a broad range of age and ability level, and 4 subtests (Alertness, Distractibility, Go/NoGo, and Flexibility) were found to be feasible for the majority (>90%) of participants with FXS. Specific scores from those subtests showed good reproducibility (ICC>0.7) and clinical validity when compared with parent ratings on standardized scales (p<0.01 for correlation with ratings of hyperactivity or attention), but it has not previously been reported whether the KiTAP can detect change due to an intervention in an FXS cohort.

**Objective:** To evaluate the KiTAP with respect to sensitivity to change in 3 sets of analyses from clinical trials of disease-targeted pharmacological interventions in FXS.

**Methods:** The KiTAP was administered at baseline and 3 months in two placebo-controlled trials of similar design studying effects of 3 months of treatment with AFQ056 (Novartis) in adolescents and adults with FXS. 45 (40M, 5F) total subjects with IQ<70 were evaluated (age 12-45) in these studies. Percent change in KiTAP performance from baseline to 3 months was compared between the AFQ056 and placebo groups in the combined dataset from the two studies. Data was also analyzed using least square mean (LS mean) differences controlling for baseline performance. The KiTAP was administered at baseline, 2 weeks, and 6 weeks in a placebo-controlled trial studying effects of 6 weeks of Metadoxine Extended Release (MDX, Alcobra) treatment in adolescents and adults with FXS. 57 (43M, 14F) subjects with IQ range 19-107 were evaluated (age 14-50). A mixed effect model for repeated measures was used to compare LS mean differences from baseline to 6 weeks between MDX and placebo groups. Data from the KiTAP were also compared in a preliminary analysis between baseline and 3 months of minocycline/placebo treatment in a placebo-controlled crossover trial studying effects of minocycline in children with FXS. 34 (26M, 8F) subjects with mean IQ=55 were evaluated (age 5-16).

\(^5\) Alcobra, Inc.
**Results:** In the AFQ056 combined study group there was no significant worsening on any measure from any of the 4 subtests administered, a significant increase in correct answers (p=0.03) and decrease in omissions (p=0.03) on Go/NoGo, and a trend toward more correct answers on Flexibility (p=0.059) for the AFQ056 group (n=32) relative to placebo (n=13). These results were confirmed using the LS mean analysis. In the MDX study, there was no significant worsening on any subtest and a significant reduction in the number of errors (p=0.043) on Go/NoGo in the MDX group (n=26) relative to placebo (n=31), and a trend towards lower median response time on Distractibility (p=0.072) in the MDX group (n=27) relative to placebo (n=31). In the minocycline study (n=19 completing KiTAP all administrations), there was a significant improvement in response time on Alertness and Distractibility (p<0.05) and variation in response time on Alertness (p<0.05) for minocycline treatment relative to placebo.

**Discussion:** The KiTAP, an executive measure that can be done successfully by a high percentage of adults and adolescents with FXS and shows good reproducibility and clinical validity, was sensitive to pharmacological intervention, particularly for impulsivity on the Go/NoGo subtest and speed of response on Distractibility, each subtest showing improvement in two separate clinical trials of disease-directed agents in FXS.

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**Paper Title:** Sensitivity of an Eye Tracking Paradigm as an Outcome Measure in a Fragile X Syndrome Targeted Treatment Clinical Trial

**Authors:** Stephanie Sansone¹, Elizabeth Berry-Kravis², Crystal Crestodina³, Yingratana McLennan¹, Randi Hagerman¹, David Hess¹

**Introduction:** Gaze avoidance is a hallmark phenotypic feature of fragile X syndrome (FXS) that reflects social anxiety, hyperarousal, and can significantly impair social reciprocity and engagement¹-³. Using an infrared eye-tracker, we previously developed a paradigm to objectively measure this aspect of the phenotype and have demonstrated that individuals with FXS make fewer gaze fixations and reduced looking time to the eye region of human faces when compared to typically developing controls². Furthermore, test-retest data from this paradigm showed that eye gaze behavior was highly reliable across a period of approximately two weeks³. However, the sensitivity of this paradigm as an outcome measure in early stage clinical trials had yet to be determined. To address this we examined eye gaze behavior using the aforementioned paradigm prior to and following 12 weeks of treatment with AFQ056 (Novartis), an mGluR5 antagonist targeted treatment for FXS.

**Methods:** Individuals participated in a randomized, double-blind, placebo-controlled clinical trial of AFQ056 in adolescents or adults diagnosed with FXS. Participants were between 12 and 45 years old, had an IQ below 70, and as part of the trial were randomly assigned to receive either one of three doses of AFQ056 (25mg, 50mg, or 100mg) or placebo. Prior to randomization subjects completed the eye tracking paradigm and at a second visit after receiving three months of treatment.

The following results are based on the analysis of a subsample of individuals who were enrolled at either the RUSH University or UC Davis sites (N = 57) where eyetracking was available. Tobii T120 infrared binocular eye trackers were used to collect gaze pattern data during a passive viewing paradigm. Stimuli were the same as those used by Farzin and colleagues²,³ which consisted of 60 color photographs of adult human faces. Stimuli had either a calm, happy, or fearful expression. Based on previous findings²,³, analyses focused on in the proportion of looking time and fixations to the eye region. Eye tracking data were assessed using a mixed-effects model with random effects for emotion and individual nested within study site.

**Results:** On average, individuals when treated with AFQ056 showed a significant increase in the proportion of time looking to the eye region compared to baseline (β = .02, p = .02). There was no significant difference between treatment and baseline for the placebo group (β = .0006, p = .96). Individuals treated with AFQ056 showed a "trend" toward an increase in the number of
fixations recorded in the eye region compared to baseline (β = .05, p = .07). We also observed a significant difference in the number of fixations in the eye region when on placebo compared to baseline, albeit in the opposite direction compared to the AFQ056 treated group (β = -1.09, p = .02).

Discussion: Results from this laboratory-based study of social gaze behavior shows that AFQ056 reduces gaze avoidance in FXS. Follow-up analyses are required to establish whether these changes are linked to clinical improvement. Our findings support this eye tracking protocol as an objective, physiologically-based, reliable and sensitive outcome measure that captures aspects of the FXS phenotype.

References/Citations:

Symposium Title: Parent and Child Outcomes Following Parent-Focused Stress Reduction, Mindfulness-Based, and Gratitude Interventions

Chair: Naomi Ekas

Discussant: Nancy Miodrag

Overview: Parents of children and adults with intellectual and developmental disabilities face unique challenges that may negatively affect their well-being. Indeed, research shows that these parents report elevated levels of stress, depression, and anxiety compared to parents of typically developing children. In recent years, studies have shown that parent-focused interventions may be successful in reducing parent stress and increasing well-being. In this symposium we present three studies that examine the efficacy of three different interventions: stress reduction, mindfulness, and gratitude.

Paper 1 of 3

Paper Title: Outcomes and Feasibility of a Gratitude Intervention for Mothers of Children with Autism Spectrum Disorder

Authors: Naomi Ekas, Lisa Timmons, Megan Pruitt, Cathy Cox

Introduction: Parents of children with autism spectrum disorder (ASD) experience increased parenting stress and depression compared to parents of typically developing children. Gratitude is an emotion, practice, or dispositional tendency of being thankful for something or someone in one's life (Watkins et al., 2003). Interventions which incorporate practices such as giving thanks can improve well-being. For example, participating in a letter-writing activity to express gratitude was associated with increased life satisfaction and subjective happiness, as well as decreased levels of depressive symptoms (Toepfer et al., 2012). Although interventions for parents of children with ASD exist, there are no studies that have examined the efficacy of gratitude-inducing intervention for this population.

Methods: Sixty-seven mothers of a child with ASD (< 18 years old) participated in this study. The procedure used for the study was based on a gratitude intervention implemented by Lyubomirsky and colleagues (2011). Participants were told they would be in a happiness intervention. After completing a pre-intervention assessment assessing well-being (e.g., depression, life satisfaction, optimism, parenting efficacy, and relationship satisfaction), participants were randomly assigned to one of three conditions: (1) general gratitude (n = 24); (2) child-specific gratitude (n = 22); or (3) control (n = 21). Participants were instructed to write for 15 minutes, once per week for a total of 8 weeks. Participants in the general gratitude group wrote a letter to someone they were grateful for. In the child-specific group they wrote a letter to their child with ASD. Those in the control group wrote a list of what they did that week. A follow-up survey was completed immediately following completion of the intervention activities and an additional follow-up was completed one month later.

Results: Repeated measures ANOVAs were conducted to examine changes from pre- to post-intervention. We found significant improvements in well-being for all participants. All groups reported a decrease in negative affect from pre- to post-intervention ($F(1, 64) = 10.46, p < .01$), parental self-efficacy increased ($F(1, 64) = 20.31, p < .001$), and parents' ratings of their child's positive contribution increased ($F(1, 64) = 10.46, p < .01$). The one month follow-up data has been collected and will be analyzed shortly. We also used linguistic software to analyze the narrative content of mothers' writing. We found that mothers in the gratitude writing conditions used more nostalgia-related words compared to the control condition. Mothers whose narratives contained more nostalgic words reported increased relationship satisfaction and optimism at the post-intervention assessment.

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Discussion: The purpose of this study was to examine the feasibility and effectiveness of a gratitude intervention for mothers of children with ASD. We found that participants in both the intervention and control groups showed improvements in well-being. Although the improvements in the control group were surprising, it could be that reflecting on the previous week's accomplishments were responsible for the improvements. Finally, the intervention was designed to induce feelings of gratitude; however, our analyses show that mothers' narratives included nostalgic elements that were associated with increases in well-being. This intervention shows promise for improving well-being and further studies with larger samples are needed.

References/Citations:

Paper 2 of 3

Paper Title: The Impact of Behavior Services and Parent Mindfulness-Based Stress Reduction on Child Behavior Problems: Implications for a Combined Treatment Model

Authors: Cameron L. Neece

Introduction: Approximately 50% of children and adolescents with intellectual and developmental delays (IDD) have serious behavior problems—a prevalence three times greater than what is seen in typically developing youth (Baker et al., 2010). Parents of these children experience alarming levels of parenting stress (Davis & Carter, 2008), and recent research suggests that parenting stress is a robust predictor of subsequent behavior problems and poor social and emotional competence, leading to later mental disorders in youth with IDD (Baker et al., 2010; C. Neece & Baker, 2008; C. L. Neece et al., 2012). Further, high parental stress predicts less beneficial outcomes for children in early intervention programs (Osborne, McHugh, Saunders, & Reed, 2008; Robbins, Dunlap, & Plienis, 1991; Strauss et al., 2012) and fewer gains in parenting skills in behavioral parenting training interventions (e.g., Baker, Landen, & Kashima, 1991). Therefore, intervening directly on parenting stress may have cascading effects on both family and child outcomes. Recent research suggests the Mindfulness-Based Stress Reduction (MBSR) is effective in reducing parental stress (Dykens et al., 2014; Minor et al., 2014; Neece, 2015; Bazzano et al., 2013) as well as improving child behavior problems (Neece, 2014) and social skills among families of children with IDD (Lewallen & Neece, 2015). Thus, interventions that address both child behavior problems (e.g. in-home behavioral services) and parent stress (e.g. MBSR) may be more effective than the interventions independently. The aim of the current study was to examine changes in parenting stress, child behavior problems, and child social skills among families where the child and parent received no intervention (NoI), the child received in-home behavioral services only (Beh-only), the parents received MBSR only (MBSR only), and families where the parent and child both received intervention (Comb).

Methods: The current study included 79 parents of children, ages 2.5 to 5 years old, with IDD. These parents participated in a randomized controlled trial examining the efficacy of MBSR in reducing parental stress and subsequent child behavior problems. Parents were assigned to an immediate treatment or a waitlist control group and data was collected on whether the child was receiving behavioral services or not. The four groups included 1) families assigned to the control group whose child was not

3 Loma Linda University
receiving behavioral services (NoI, N=25), 2) families assigned to the control group where the child was receiving behavioral intervention (Beh-only, N=20), 3) families assigned to the treatment group whose children were not receiving behavioral services (MBSR-only, N=15), and 4) families assigned to the treatment groups whose while were receiving behavioral services (Comb, N=19). Parenting stress was measured using the Parenting Stress Index (Abidin, 1990), child behavior problems were assessed with the Child Behavior Checklist (Achenbach & Rescorla, 2000), and child social skills were evaluated using the Social Skills Improvement System (Gresham, Elliott, Cook, Vance, & Kettler, 2010)

Results: Results indicated that there were overall group differences in improvements in parental stress, child ADHD symptoms, child attention problems, and child empathy. Regarding parental stress, parents in the Comb group reported lower scores post-treatment compared to parents in the NoI and Beh-only groups (F=7.02, p<.001). In terms of child outcomes, children in the Beh-only group had significantly higher ADHD symptoms and attention problems and significantly lower Empathy scores following treatment compared to the MBSR-only group (ADHD F=3.85, p<.05, Attention F=3.48, p<.05, Empathy F=2.83, p<.05). Future analyses will investigate potential moderators of the observed relationships (e.g. number of hours of behavioral services, satisfaction with behavioral services) as well as examine teacher reports of changes in child behavior problems and social skills across the four groups.

Discussion: These findings highlight the importance of targeting parents stress in interventions designed to improve child behavior and social outcome as well as suggest the possible benefits of using a combined approach. Other clinical implications and directions for future research will be discussed.

Paper 3 of 3

Paper Title: Mindfulness-Based Intervention for Parents of Adults with Intellectual / Developmental Disabilities: Outcomes from a Randomized Active Treatment Controlled Trial

Authors: Yona Lunsky4, Richard Hastings5, S. Hutton6, C. McMorris7, A. Palucka4, J. Weiss7, K. White8

Introduction: Despite the number of recent studies focused on mindfulness based interventions for parents of people with intellectual / developmental disabilities (IDD), only 2 studies have included active treatment control groups. Both have focused either exclusively (Ferroli & Harris, 2013) or primarily (Dykens et al. 2014) on parents of children and youth. The purpose of the current study was to compare clinical outcomes for parents of adult children with IDD randomly assigned to either a mindfulness based or support and education focused parent intervention.

Methods: Out of approximately 900 parents seeking adult services for their children age 16 and up with IDD, 138 parents expressed interest in the intervention project. Eligible participants were randomly assigned to one of two parent groups, 2 hours weekly for 7 weeks. The mindfulness based intervention group was developed for parents of adults with IDD (Lunsky et al., 2015) The support and education intervention group had a psychoeducational component followed by group discussion. Fifty parents completed baseline measures, with 26 randomly assigned to mindfulness and 24 to support and information. Twenty-six parents in the mindfulness group and 20 parents in support and information completed post-intervention ratings, and 20 parents from mindfulness and 17 parents in support and information completed three month follow-up ratings.

4 Centre for Addiction and Mental Health
5 University of Warwick
6 Community Living Toronto
7 York University
8 DSO Toronto
Parent psychopathology was measured by the Depression, and Stress subscales from the DASS-21, and mindfulness was measured by the Five Facets of Mindfulness Questionnaire (FFMQ) and the Bangor Mindful Parenting Scale (BMPS).

**Results:** Analysis of covariance indicated a significant main effect of group assignment on depression at Time 2, (F (1, 43)=11.99, p=.001), and on stress at Time 2 (F (1, 43)=6.63, p=.04), when adjusting for Time 1 scores. In both cases, the reductions in distress were in favor of mindfulness. There was no significant main effect of group assignment on the FFMQ or BMPS.

At follow-up, a repeated measures ANCOVA revealed no significant group x time interactions or within subject effects in DASS-Depression or Stress scores, suggesting no change between post intervention and follow-up for either group. This provided evidence of the maintenance of the initial intervention gains.

**Discussion:** Using a robust, randomized, attention-controlled design our data suggest a benefit of mindfulness based intervention for parents of adults with IDD. These are the first more tightly controlled data on the use of mindfulness with parents of adults. Initial benefits were maintained over a short follow-up period. The study has also highlighted important feasibility data, including the challenges of recruiting parents of adults with IDD across a large metropolitan area. Further research needs to include a large scale multi-site definitive effectiveness trial, examination of long term maintenance of effects, and exploration of alternatives to face-to-face interventions for parents who are harder to reach.

**References/Citations:**

Symposium Title: Can Rare Disorders Pave the Way to Targeted Interventions for Autism Spectrum Disorders (ASD) and Intellectual Disability (ID)? Insights Gained from Fragile X Syndrome, Tuberous Sclerosis Complex, and Dup15q Syndrome

Chair: Shafali S. Jeste1 2

Discussant: Julian Martinez1

Overview: Precipitated by rapid advances in diagnostic methods, from the chromosomal microarray to whole exome sequencing, routine clinical genetic testing is now recommended for the etiological evaluation of all children with new diagnoses of neurodevelopmental disorders, including global developmental delay, Intellectual Disability (ID) and Autism Spectrum Disorder (ASD) (Shevell et al, 2003; de Vries et al., 2005; Schaefer et al, 2013; Moeschler et al, 2014). This surge in genetic testing has facilitated the identification of causative rare genetic variants and, with the ascertainment of subgroups of individuals with shared variants, the identification of clinically meaningful genetic syndromes (Jeste and Geschwind, 2014). To date, detailed developmental and behavioral characterization of these syndromes has lagged behind the genetic diagnoses, leaving considerable uncertainty at the time of diagnosis regarding developmental trajectories, prognosis and recommended treatment options. Interventions remain broad in focus, targeting the neurodevelopmental diagnoses (such as ASD or ID) rather than specific features of cognition or social communication that may define a particular syndrome. This considerable dissociation between the precision of genetic testing and the imprecision of clinical treatment may be addressable and represents a critical challenge in neurodevelopmental disorders. Improved and precise behavioral characterization of genetic syndromes associated with ID and ASD can inform not only prognosis, but also treatment, with the ultimate goal of facilitating the discovery of targeted, mechanism-based interventions that may improve individual outcomes.

Paper 1 of 3

Paper Title: The Comorbidity of Fragile X Syndrome and Autism Spectrum Disorder: Failed and Potential Insights

Authors: Len Abbeduto3 4, Andrea McDuffie3 4, Angela J. Thurman3 4

Introduction: More than half of individuals with fragile X syndrome (FXS) also meet diagnostic criteria for autism spectrum disorder (ASD). This high rate of comorbidity has led to claims that because FXS is an etiologically "simpler" (i.e., a single-gene) disorder, it provides a ready window into the more etiologically complex nonsyndromic ASD (Belmonte & Bourgeron, 2006). Moreover, the high comorbidity has led to the claim that targeted pharmaceutical treatments that are efficacious for core symptoms of FXS are likely to be beneficial for nonsyndromic ASD as well (Berry-Kravis et al., 2012). Arguably, however, few insights into nonsyndromic ASD have yet to come from research on FXS, and no pharmaceutical treatments developed for FXS have had documented benefit for core symptoms of ASD. In part, the failure of research on FXS to advance the understanding and treatment of ASD reflects the fact that although the comorbidity of the two disorders was first recognized over 30 years ago (Brown et al., 1982), there is still much that we do not understand about the comorbidity. In fact, many of the claims about the potential for synergy between research on FXS and nonsyndromic ASD derive from the assumption that ASD symptoms reflect the same underlying psychological and neurobiological impairments in both disorders.

Methods: In the proposed presentation, we will argue that this assumption is probably not correct. We will present evidence from published and unpublished findings from our laboratory that strongly suggest that ASD symptoms in FXS and in

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nonsyndromic ASD reflect at least partly different underlying impairments. We will summarize four sets of findings relevant to understanding the FXS-ASD comorbidity.

Results: First, we will present data on the symptoms that lead to the diagnosis of ASD in FXS and the nonsyndromic case, thereby providing evidence of differences in the symptom profiles "earning" affected individuals the diagnosis. Second, we will present data on behaviors and impairments that, although not diagnostic of ASD, are nonetheless correlated with the diagnosis or correlated with ASD symptom severity, which again point to important differences between FXS and nonsyndromic ASD. Third, we will present data on differences in the developmental trajectories of ASD symptoms in FXS and nonsyndromic ASD. Fourth, we will present data on the symptoms and correlates that distinguish individuals with FXS who receive an ASD diagnosis from those with FXS who do not receive the diagnosis.

Discussion: Together, these data suggest that although there are similarities between the ASD symptoms and profiles of FXS and nonsyndromic ASD, there are numerous clinically important differences as well. We will conclude the presentation by suggesting that use of the categorical diagnosis of ASD in FXS obscures important differences within the syndrome and between the syndrome and nonsyndromic ASD and is thus, a barrier to scientific progress and to the development of effective treatments. Instead, we favor focusing on individual symptoms or constellations of symptoms in future research, including studies designed to evaluate treatment efficacy.

References/Citations:


Paper 2 of 3

Paper Title: Characterizing Early Developmental Trajectories and Social Communication Profiles in Tuberous Sclerosis Complex

Authors: Kandice Varcin5 6, Shafali S. Jeste1 2, Amanda Gulsrud1 2, Charles Nelson5 6

Introduction: Tuberous Sclerosis Complex (TSC) is an autosomal dominant genetic disorder resulting from a TSC1/TSC2 mutation. This genetic disorder confers a high risk for neurodevelopmental disorders, including autism spectrum disorder (ASD) and intellectual disability (ID) (Curatolo, 2015). Up to 80% of children with TSC will experience cognitive delay and up to 60% will meet criteria for ASD (Jeste, 2008). Importantly, ID and ASD often co-occur in this population, posing challenges for diagnosis, intervention targets and treatment selection. The high rate of ASD and ID, combined with the fact that TSC is often diagnosed prior to the onset of any social communication or cognitive delays, has led to TSC being studied as a "model system" for neurodevelopmental disorders in both animal and human research. However, despite hypotheses about the role of epilepsy, cortical pathology, and co-occurring genetic mutations in predicting neurodevelopmental outcomes in TSC, no single clinical factor has been identified as a consistent predictor of atypical neurodevelopment. Moreover, beyond prevalence rates, we know relatively little about the cognitive and phenotypic profiles of children with TSC and the degree to which these profiles show

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convergence with non-syndromic ASD. In this context, we recently conducted the first prospective, longitudinal study of early development in infants with TSC (Jeste, 2014). We aimed to (1) characterize early developmental trajectories in TSC, (2) identify prospective risk factors of ASD and ID that would allow us to define the timing and emergence of developmental delays, and (3) compare cognitive and behavioral profiles in TSC to non-syndromic ASD and typical development. Collectively, we aimed to move beyond diagnostic categorization to a deeper and more fine-grained characterization of early development in this high-risk group in order to inform intervention targets.

Methods: Infants with TSC were recruited as early as 3-6 months of age and followed longitudinally until 36 months of age. We took a multi-modal approach to characterize behavioral, neurophysiological and clinical changes in early development. Data gathered at each time point included detailed seizure history, developmental testing using the Mullen Scales of Early Learning (MSEL), social-communication assessments including the Autism Observation Scale of Infancy (at 6, 9 and 12 months) and the Autism Diagnostic Observation Schedule (ADOS) at 18, 24 and 36 months. We also collected eye tracking data in response to faces and electroencephalographic (EEG) data in baseline and event-related conditions.

Results: Infants with TSC show delays in cognitive development and social communication skills, particularly in the non-verbal domain, as early as 6 months of age. Infants with TSC who develop ASD show the greatest cognitive impairment from 12 months age and a slowing of non-verbal skills development between 12 to 36 months of age compared to infants with TSC without ASD (Jeste et al., 2014). Using EEG, we have demonstrated that non-verbal delays do not appear to be rooted in deficits in low-level visual processing and may instead represent impairments in networks associated with higher-level cognitive processes (Varcin, 2015). Through a profile analysis, comparing individual item-scores on the ADOS, we have identified a pattern of impairment in social communication and restrictive and repetitive behaviors in toddlers with TSC and ASD that closely maps that of toddlers with non-syndromic ASD.

Discussion: Our research to date has unveiled a developmental profile in TSC that is marked by prominent and early delays in non-verbal skills, particularly in infants with TSC and ASD. Despite the prominence of intellectual disability and seizures, infants with TSC and ASD show a profile of social communication impairment that has almost complete convergence to that of children with non-syndromic ASD. These results have laid the foundation for the first trial of early behavioral intervention in TSC.

References/Citations:

Paper 3 of 3

**Paper Title:** Recent Advances in 15q11.2-q13.1 Duplications (Dup15q Syndrome): Identification of Symptom Profiles Within the ID and ASD Spectrum

**Authors:** Shafali S. Jeste, Charlotte DiStefano, Amanda Gulsrud, Larry Reiter, Ronald Thibert, Edwin Cook

**Introduction:** Several chromosomal disorders, copy number variant (CNV) syndromes and single nucleotide variants have been associated with autism spectrum disorder (ASD) and Intellectual Disability (ID), with the number increasing recently at a relatively rapid rate (Schaefer, 2013; Moeschler, 2014). Duplications of the 15q11.2-q13 region of maternal origin (Dup15q syndrome) were first associated with ASD and ID over 15 years ago, and now these duplications are amongst the most common CNV’s associated with ASD and related neurodevelopmental disorders (Hogart, 2010). Through collaborative efforts facilitated by a national Dup15q alliance, studies have identified neurobiological, developmental and behavioral features of Dup15q syndrome that will guide targeted treatments, both behavioral and pharmacological. In the last year, we have been studying the behavioral and electrophysiological characteristics of children with Dup15q syndrome compared to IQ matched children with non-syndromic ASD in order to determine if there are distinctive features of the social-communication deficits and overall developmental profiles of children with Dup15q syndrome. Our study is driven by the hypothesis that there will be a distinctive developmental phenotype in children with Dup15q syndrome that will vary in severity across mutation size and location, and that this phenotype will be defined by expressive language and motor impairment with relatively preserved non-verbal social abilities. We frame these findings in the context of larger, retrospective studies of the clinical symptom profile of children with Dup15q syndrome.

**Methods:** The Dup15q Alliance has been collecting a registry of patients, with 425 patients in the registry and 212 with complete clinical records. From the registry, 142 children have isodicentric 15q duplications, 30 have interstitial duplications, 14 have mosaic isodicentric duplications, and 26 have "edge" duplications either at q11.2 or q13.3. We recruited a cohort of children from the UCLA Dup15q clinic for a comprehensive, developmentally targeted behavioral evaluation and compared them to an IQ and age matched cohort of children with ASD. Participants were assessed for verbal and non-verbal cognition, ASD characteristics based on the Autism Diagnostic Observation Schedule (ADOS) and adaptive function based on the Vineland Adaptive Behavior Scales (VABS). Group comparisons were performed between Dup15q and ASD participants, as well as within the Dup15q group based on duplication type and epilepsy status.

**Results:** Participants included 13 children with Dup15q syndrome and 13 children with non-syndromic ASD, ages 22 months - 12 years. Of the children with Dup15q syndrome, 10 participants had isodicentric and 3 had interstitial duplications. Four children had active epilepsy (all with isodicentric duplications). All children with Dup15q syndrome met criteria for ASD, but ASD severity scores were significantly lower than children in the non-syndromic ASD group. ADOS profiles demonstrated a relative strength in items related to social interest. Children with Dup15q syndrome also demonstrated significantly more impairment in motor and daily living skills. Within the Dup15q group, children with epilepsy demonstrated significantly lower cognitive and adaptive function than those without epilepsy.

**Conclusions:** The relative strength observed in social interest and responsiveness in the context of impaired motor skills represents an important avenue for intervention, including aggressive treatment of epilepsy, early and consistent focus on motor skills, and intervention targeting joint attention and language within a play context, in order to build on social interest to further develop social communication abilities. Longitudinal research beginning in early development will elucidate the temporal relationships between developmental domains and neurological comorbidities in these children at high risk for

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neurodevelopmental disorders. Translational research, linking electrophysiological and behavioral phenotypes in mouse models to patients will facilitate the identification of the specific genetic mechanisms underlying the neurodevelopmental symptom profile in children with Dup15q syndrome.

References/Citations:

Symposium Title: Executive Functions in Autism Spectrum Disorder, Down Syndrome, and Sex Chromosome Aneuploidy: An Examination of Age Effects and Relations with Comorbid Psychopathology

Chair: Nancy R. Lee¹, Gregory L. Wallace³

Discussant: Deborah Fidler⁴

Overview: Executive functions (EF) are important for the completion of everyday tasks that involve planning, self-monitoring, organization, and the active maintenance of information in pursuit of a larger goal. Research suggests that virtually all developmental disabilities can be characterized by some degree of executive dysfunction, and it has been proposed that different developmental disorders can be characterized by different executive function profiles [1,2]. The proposed symposium seeks to integrate research on everyday executive functions, as measured by the Behavior Rating Inventory of Executive Function, in youth with autism spectrum disorder, Down syndrome, and sex chromosome aneuploidy paying particular attention to the following: (a) variation in EF skills as a function of age and (b) links with comorbid psychopathology.

Paper 1 of 4

Paper Title: Age and Co-Morbid Psychopathology are Associated with Everyday Executive Functioning among Children and Adolescents with Autism Spectrum Disorder

Authors: Emily White², Gregory L. Wallace³, Cara Pugliese⁵, Laura Anthony⁵, Alex Martin², Lauren Kenworthy⁵

Introduction: It is now well established that executive functioning (EF) is developmentally dynamic during typical development, with variable trajectories depending upon the subcomponent EF process under investigation (e.g., working memory vs. inhibitory control vs. cognitive flexibility). EF difficulties are also associated with various forms of psychopathology, including anxiety, depression, and aggression. Among individuals with autism spectrum disorder (ASD) relatively little is known about how EF skills in everyday settings vary at different ages and relate to co-morbid psychopathology. The first aim of the current study is to extend prior work by evaluating age-related differences in parent-reported EF problems during childhood and adolescence in a large cross-sectional cohort of children with ASD. The second aim of the present investigation is to examine associations between these everyday EF problems and co-morbid psychopathology.

Methods: Children (n=210; 174 males) with an ASD without intellectual disability participated in the study and each had everyday EF and co-morbid psychopathology rated by parents utilizing the Behavior Rating Inventory of Executive Function (BRIEF) and the Child Behavior Checklist (CBCL), respectively. Participants were divided into four groups based on age (5-7, 8-10, 11-13, and 14-18 year olds).

Results: There were significant age effects (i.e., worsening scores with increasing age) on three BRIEF scales: Inhibit (p=.015), Initiate (p=.015), and Monitor (p=.019). In addition, when submitted to a regression accounting for age effects, both behavior regulation and metacognitive EF problems (from the BRIEF) predicted (p<.01) depression symptomatology. In contrast, age and behavior regulation problems predicted anxiety symptomatology, and behavior regulation problems alone predicted conduct problems (ps<.01).

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Discussion: Older children with ASD show some evidence for greater EF problems compared with the normative sample than younger children with ASD. Specifically, there is a widening divergence from the normative sample in select EF subcomponents in children with ASD as they age. There were also strong associations between EF problems and co-morbid psychopathology (i.e., anxiety symptoms, depression symptoms, and conduct problems). These findings have implications for the challenges faced by individuals with ASD without intellectual disability as they attempt to enter mainstream occupational and social environments. Much work is still needed. The gold standard approach for assessing age effects is the utility of a longitudinal design. We are currently in the final stages of collecting follow-up BRIEF data in a relatively large proportion of these children and adolescents with ASD. Therefore, by the time of the conference, we also aim to profile for the first time longitudinal changes in everyday EF and their correlates in ASD.

Paper 2 of 4

Paper Title: Real-World Executive Functioning and its Clinical Correlates among Young Adults with Autism and Older Adults with the Broader Autism Phenotype

Authors: Gregory L. Wallace\(^3\), Rebecca Charlton\(^6\), Haroon Popal\(^7\), Emily White\(^3\), Jessica Budgett\(^6\), Lauren Kenworthy\(^5\), Alex Martin\(^2\)

Introduction: Although executive functioning (EF) difficulties are well documented among children and adolescents with autism spectrum disorder (ASD), little is known about real-world measures of EF among adults with ASD. Therefore, we present two studies of real-world EF and its associations with co-morbid psychopathology in (1) young adults with ASD and (2) older adults with the Broad Autism Phenotype (BAP).

Methods: Study 1 examined parent-reported real-world EF problems (from the Behavior Rating Inventory of Executive Function-Adult version) among 35 adults (ages 18-40 years) with ASD without intellectual disability and how they correlated with co-morbid anxiety and depression symptomatology (using the Adult Behavior Checklist) and with adaptive functioning (using the Adaptive Behavior Assessment System-Second Edition). Study 2 investigated for the first time the BAP (using the Broad Autism Phenotype Questionnaire) in the context of older adulthood and its associations with real-world executive function (from the Behavior Rating Inventory of Executive Function-Adult version), social support (Duke Social Support Index), and both depression (Geriatric Depression Scale) and anxiety (Beck Anxiety Inventory) symptomatology. Based on self-ratings of autistic traits and the presence of family members with an autism spectrum disorder diagnosis, 66 older adults (60+ years old, range=61-88) were split into BAP (n=24) and control (n=42) groups.

Results: In study 1, a variable EF profile was found with the most prominent deficits occurring in flexibility and metacognition (ps<.001). Flexibility problems were associated with anxiety-related symptoms (p<.01) while metacognition difficulties were associated with depression symptoms and impaired adaptive functioning (ps<.05). In study 2, individuals in the BAP group, even after controlling for age, education level, sex, and health problems (e.g., high blood pressure, diabetes) exhibited more real-world executive function problems in multiple domains (with a profile akin to prior studies of children [and to the study described above of adults] with ASD, including a peak deficit in flexibility), reported lower levels of social support, and self-rated increased depression and anxiety symptomatology compared to the control group (ps<.001). Additionally, just as in study 1, flexibility problems were associated with anxiety-related symptoms (p<.001) while metacognition difficulties (as well as flexibility problems in this case) were associated with depression symptoms (ps<.01).

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Discussion: Study 1 revealed that real-world EF problems persist into adulthood in ASD and are strongly associated with both internalizing behaviors and adaptive functioning making EF an important treatment target among adults with ASD. Findings from study 2 suggest that the BAP in older adulthood could represent an additive risk factor in which normal age-related declines are exacerbated by the presence of increased autistic traits. These results might in turn inform our understanding of aging in autism spectrum disorder, which has been largely unexplored to date. These two studies converge in demonstrating the continued importance of everyday EF throughout adulthood in ASD and the BAP. Just as studies of EF across childhood and adolescence in ASD suggest persistent flexibility difficulties, so too do these studies of young adults with ASD and older adults with the BAP. Moreover, both studies converge in demonstrating an emerging link between EF problems and psychopathology, specifically flexibility problems with anxiety symptoms and metacognition problems with depression symptoms. Given the relatively new EF-based interventions for ASD in childhood, it is imperative to extend these upward developmentally as they may improve outcomes for adults with ASD.

Paper 3 of 4

Paper Title: An Examination of The Relations Between Everyday Executive Functions, Age, and Psychopathology in Children and Young Adults with Down Syndrome

Authors: Mary Godfrey1, Moshe Maiman1, Payal Anand3, Elizabeth Will4, Liv S. Clasen7, Lisa Daunhauer4, Deborah Fidler4, Jay Giedd2,8, Jamie Edgin7, Nancy R. Lee12

Introduction: Down syndrome (DS) is a developmental disorder characterized by a slowing of cognitive development from infancy to very early childhood [1,2], a precocious decline in executive function [EF] skills in later adulthood [3], and heightened rates of early onset Alzheimer’s disease [4]. Less is known about the relationships between age and different cognitive abilities, such as EF skills, in childhood and young adulthood in this group. Thus, the current research sought to describe age-EF relations in a sample of individuals with DS, ages 4-24 years. In addition, it aimed to examine the behavioral correlates of parent-reported EF skills by examining relations between scores on the Behavior Rating Inventory of Executive Function (BRIEF) and dimensions of psychopathology as measured by the Nisonger Child Behavior Rating Form (NCBRF).

Methods: The sample included 85 youth with DS and 43 youth with typical development (TD), ages 4-24 years. Parents completed either the preschool or school-age version of the BRIEF and raw composite scores were calculated for five domains (Emotional Control, Inhibition, Shift, Working Memory, and Plan/Organize) by identifying identical or very similar items across the two forms of the BRIEF. In addition to completing the BRIEF, a subset of parents (n=22) also completed the NCBRF to measure different domains of psychopathology in youth with DS only.

Results: To examine if age-BRIEF relations varied as a function of group, hierarchical linear regression analyses were completed with the following steps: (1) age, (2) group, (3) age x group. Results revealed main effects of group across all composites (all ps < .01), main effects of age on the Inhibit composite (p<.05), but no group x age interactions (ps>.4). Rather, similar degrees of impairment were observed for youth with DS relative to the TD group over the age range studied for all five BRIEF domains. Relationships between BRIEF scores and dimensions of psychopathology/prosocial behavior were examined using stepwise linear regression (forward selection) to predict the following NCBRF scales: Conduct Problem, Insecure/Anxious, Self-Isolated/Ritualistic, Compliant/Calm, and Adaptive Social. (These scales were chosen in order to minimize item overlap between the BRIEF and NCBRF). For the Conduct Problem scale, the BRIEF Inhibit and Plan/Organize composites were significantly predictive of problem behaviors (all ps < .05). For the Insecure/Anxious and Self-Isolated/Ritualistic scales, only the Emotional

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Control composite of the BRIEF was significantly predictive (ps<.05). For both the Compliant/Calm and Adaptive Social scales, only the Inhibit composite was significantly predictive (p<.05).

**Discussion:** These results suggest that difficulties on everyday EF tasks (as rated by parents) are similar in degree across middle childhood and young adulthood in DS. Moreover, a preliminary examination of EF-psychopathology relations suggested that parent ratings of prosocial and problem behaviors in youth with DS were related to everyday EF skills falling into the Plan/Organize, Emotional Control, and Inhibit domains. These results will be discussed within the context of the larger DS cognitive-behavioral phenotype and the possible importance of EF skills for adaptive behaviors and psychopathology. Lastly, limitations of this research, including the cross-sectional sample and use of parent ratings of EF and psychopathology only, will be discussed with regard to the need for future research.

**References/Citations:**

**Paper 4 of 4**

**Paper Title:** Variation in The Severity of Everyday Executive Function Skills in Relation to Age and Symptoms of Psychopathology in Youth With an Extra X Chromosome: A Comparison with Typical Youth and Youth with Down Syndrome

**Authors:** Nancy R. Lee1,2, Elizabeth Will4, Elizabeth Adeyemi2, Liv S. Clasen2, Jonathan Blumenthal2, Lisa Daunhauer4, Deborah Fidler4, Jay Giedd3,6, Jamie Edgin7

**Introduction:** Executive functions (EF) are thought to be impaired in youth with sex chromosome trisomy (Klinefelter and Trisomy X syndromes; +1X; [1]). However, little is known about developmental trajectories associated with EF difficulties in this group or the relations between everyday EF skills and psychopathology. Research studies using lab-based measures of EF have suggested a relationship between these abilities and both attention-deficit/hyperactivity disorder [2] and autism spectrum disorder symptomatology [3]. However, these relations have not been explored using a general measure of psychopathology that assesses emotional and behavioral difficulties across many domains. Thus, the current research examines this relationship as well as age-EF relations in youth with +1X relative to youth with Down syndrome and those with typical development (TD).

**Methods:** The sample included 30 youth with +1X, 30 youth with DS, and 30 youth with TD matched group-wise on age, gender, and maternal education levels. Parents of all participants completed the school-age Behavior Rating Inventory of Executive Function (BRIEF). In addition, a subset (n=24) of parents of youth with +1X also completed the Achenbach Child Behavior Checklist (CBCL) about their child.

**Results:** To examine if age-BRIEF relations varied as a function of group, hierarchical linear regression analyses were completed with the following steps: (1) age, (2) group, (3) age x group. Results revealed main effects of group across all scales (all ps < .01) and a main effect of age on the Inhibit scale, such that scores improved with age overall (p<.05). However, for the Initiate and Plan/Organize scales, the main effect of group was qualified by a group x age interaction (ps<.03), such that there was a trend in the data for higher scores on the BRIEF (denoting greater difficulties) with increasing age in youth with +1X only. Relationships between BRIEF scores and dimensions of psychopathology were examined using stepwise linear regression (forward selection) to predict scores on the different scales of the CBCL. Relationships between the BRIEF scales and three CBCL scales (of 8 scales...
total) were noted. These were for Social (predicted by Monitor and Inhibit, ps<.05), Attention (predicted by Initiate only, p<.05), and Delinquent Behavior/Rule Breaking (predicted by Initiate only, p<.05).

Discussion: Unlike youth with DS, there is a suggestion that certain everyday EF skills (initiation and planning/organization) are more deviant from peers at older ages for youth with +1X. Given the cross-sectional nature of these data, these results must be interpreted with caution. However, from both a clinical and developmental perspective, it will be important to investigate these findings further, ideally using a longitudinal study design, in order to determine if greater attention should be paid to EF difficulties in adolescents with additional X chromosomes. Interestingly, relations between parent reports of EF difficulties and problem behaviors were very consistent with prior research utilizing laboratory EF measures in males with +1X. In particular, difficulties with monitoring and inhibition appeared to be related to social difficulties and difficulties with initiation appeared to be related to attentional difficulties and delinquent behavior. These results will be discussed in relation to the broader cognitive-behavioral and psychosocial phenotype for males and females with +1X, paying particular attention to disentangling executive dysfunction, externalizing behavior, and social impairments that are known to characterize males and females in this group.

References/Citations:
Symposium Title: The Effects of Caregiver-Associated Stress on Health-Related Outcomes and Parent-Directed Interventions

Chair: Stephanie Sansone, Nikko Da Paz

Discussant: Marsha Mailick

Overview: Extensive evidence exists documenting the ways in which caregiver-associated chronic stress can significantly affect mental and physical well-being. In general, chronic stress is associated with multiple mental and physical health issues, including increased symptoms of anxiety, depression, chronic inflammation, and immune-mediated diseases. The probability of being affected by chronic stress due to increased caregiver burden is much higher among parents caring for a child with a neurodevelopmental disorder. Collectively, caretaking challenges and distress associated with behavior problems, the stress of navigating services of their child, and a variety of other factors could have significant negative effects on the family as a whole. Fortunately, some studies examining the impact of caring for a child with neurodevelopmental disorder on maternal and family well-being have demonstrated positive adaptation and quality of life. The proposed symposium focuses on how parental stress can impact physiological changes, mental and physical health, as well as adherence to parent-directed intervention. We will discuss what differentiates those who are negatively impacted by parental stress and those who appear to be coping well.

We will begin with a presentation on how perceived stress relates to parental perceptions of health and a biomarker of stress among parents caring for an individual with autism spectrum disorder (ASD). Next, we will present a study in which a gene-environment interaction approach was used to examine the increased susceptibility for some women with the fragile X premutation to display altered HPA-axis and immune system activity with special focus on chronic stress among women who are mothers caring for a son or daughter with fragile x syndrome (FXS). Finally, we will end with a presentation on how stress associated with child behaviors and parental distress can impact adherence in a parent-directed cognitive training intervention for children with FXS.

Each of these speakers will be presenting findings that have important research and clinical implications. Understanding the factors that contribute to risk and resilience can help clinicians and families work together on ways to reduce or even prevent some of the negative health outcomes associated with chronic stress. Furthermore, researchers and clinicians alike should consider how parental stress can impact the overall effectiveness of interventions focused on the child.

Paper 1 of 3

Paper Title: Associations of Illness Perceptions and Subjective Stress with Health-Related Quality of Life and Salivary Cortisol Levels in Caregivers of Children with Autism Spectrum Disorder

Authors: Nikko Da Paz, Jan Wallander, Jitske Tiemensma

Introduction: Parents of children with autism spectrum disorders (ASD caregivers) experience poorer health and greater stress than parents in the general population. Traditionally, subjective reports provided evidence for these deleterious outcomes. Recent investigations of salivary cortisol have elucidated the physiology of caregiving-induced stress for ASD caregivers. Although factors predictive of negative health consequences have been extensively investigated, less is known about the role of illness perceptions and perceived stress in self-reported health and stress biomarkers for ASD caregivers. We hypothesize that an increase in negative perceptions will demonstrate an inverse association with subjective and objective measures of health.

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3 Waisman Center, University of Wisconsin-Madison
Methods: In this ongoing study, we currently have complete date from a sample of 35 ASD caregivers (ages 23 to 62 years; M = 38.0, SD = 10.35). Self-reported measures included the Brief Illness Perception Questionnaire (BIPQ), Perceived Stress Scale (PSS), and SF-36 health survey (HRQOL). The BIPQ assessed caregivers’ perception of their child’s autism with higher scores indicating a negative illness perception. To capture biomarkers of stress, we collected caregiver salivary cortisol at waking and 30 minutes post waking on two consecutive weekdays. Normal functioning of the hypothalamic-pituitary-adrenal (HPA) axis is characterized by a rise in concentration of cortisol during the first 30 minutes of waking, the cortisol awakening response (CAR). We calculated CAR as the average cortisol increase across both days. Bivariate Pearson correlations were conducted to evaluate relationships between illness perceptions, perceived stress, global health, and cortisol reactivity. Regression analyses were conducted separately to assess the predictive ability of illness perceptions and perceived stress on both HRQOL and CAR.

Results: Greater perceived stress significantly predicted impaired HRQOL (F(1,34) = 13.30, p = .001, r2 = .29) and CAR levels (F(1,34) = 6.38, p = .017, r2 = .16). Models regressing illness perceptions on HRQOL and CAR did not present significance results. ASD caregivers who reported greater stress experienced reductions to the trajectory of morning cortisol, which may indicate elevated cortisol at waking. Normal levels of CAR present increases of 2.5 nmol/L for populations with low stress. However, the mean CAR for this sample of ASD caregivers was larger (M = 3.55, SD = 2.94), confirming a postulation of elevated levels and supporting the inference of dysregulation of the HPA axis.

Discussion: Although illness perceptions did not demonstrate significant associations with unfavorable health outcomes, results revealed a significant positive association with subjective stress. Quite possibly targeting a reduction in caregiver’s negative perceptions of their child’s autism might reduce perceived stress which may, in turn, attenuate the negative consequences of poor health as indicated by self-report and irregular cortisol activity. Upon replication, it would be advantageous to consider these results when designing interventions targeting mental and physical well-being of caregivers parenting children with ASD.

References/Citations:
findings is the increased FMR1 transcription and possible toxic-gain-in-function associated with alleles in this range. Another less commonly studied hypothesis proposes that these same outcomes are also related to the increased probability of being a caregiver in this population and the consequences of coping with the chronic stress that accompanies such a role. To reconcile these seemingly opposing views, we used a gene x environment interaction approach to determine the extent to which FMR1 genotype and chronic stress interact to differentially and collectively affect HPA-axis and immune system functioning.

**Methods**: Seventy-five women (aged 19-55 years; M = 34.45, SD = 8.80) completed multiple health- and stress-related questionnaires, provide blood samples, and were given a salivary cortisol home sample kit. Blood samples were used to quantify FMR1-specific biological information and circulating cytokine levels. Forty-eight women had CGG-repeat sizes in the premutation range, of these 21 had at least one son/daughter with fragile X syndrome (FXS) and 29 had no children affected by fragile X or any other disability.

**Results**: Results from multilevel modeling analyses revealed that women with activation ratio-corrected CGG (AR-CGG) repeat lengths greater than 80 who experience greater levels of stress display attenuated diurnal cortisol trajectories and elevated waking cortisol levels. Furthermore, waking cortisol was associated with stress and AR-CGG repeat length in a curvilinear matter, such that women with AR-CGG repeat lengths between 60 and 90 repeats were more sensitive to the effects of stress. Estimates from Tobit regression models revealed a significant association between FMR1 genotype and interleukin-10 (IL-10) that was moderated by levels of stress and a trend for interleukin-12p70 (IL-12p70). Specifically, women with FMR1 alleles in the mid-premutation range displayed higher IL-10 and lower IL-12p70 with increasing levels of stress.

**Discussion**: These findings support current theory linking elevated stress with changes in the Th1/Th2 immune response balance as well as findings of increased susceptibility to HPA-axis and immune system dysfunction among women with FMR1 in the mid-premutation range. In sum, it appears that under conditions of elevated stress women with mid-size FMR1 permutation alleles are more likely to display elevated waking cortisol, flatter diurnal cortisol trajectories, and lower levels of Th1 immune mediators. Together these findings have implications for understanding the association between the FMR1 gene and the increased risk of developing depression, anxiety, autoimmune disease and various negative health outcomes among women with the premutation.

**References/Citations**:

**Paper Title**: The Impact of Parent Stress on Cognitive Training with Individuals with Fragile X Syndrome

**Authors**: Yingratana McLennan¹, Stephanie Sansone¹, Cindy Johnston¹, David Hess² ³

**Introduction**: Children with developmental disabilities benefit from a responsive parenting style in which the parent follows the child's focus of attention (Siller and Sigman, 2002). Conversely, existing studies have demonstrated a negative impact of caregiver distress on children's wellbeing. Stress and low self-esteem in mothers have been linked with less beneficial outcomes in early intervention programs (O'Connor, T.G. 2002). In a clinical trial of working memory training facilitated by caregivers for
children with fragile X syndrome (FXS), we predicted that parenting stress is associated with treatment adherence, regardless of child cognitive improvements during the therapy.

**Methods:** In this randomized, controlled study, children are assigned to adaptive or non-adaptive Cogmed (Pearson Education, Inc.) working memory training, spanning 25 sessions, 5 days per week for 5 continuous weeks. The training sessions aim to improve working memory, attention, and behavior in children and adolescents with FXS. Depending on baseline cognitive aptitude, children either trained for 15 minutes (Cogmed JM) or 30 minutes (Cogmed RM) per session while their caregiver (often mother) supervised to ensure compliance and attention to the training protocol. In addition, participants were further divided into an adaptive group in which trainings 'adapt' (become more challenging) and non-adaptive group in which trainings did not increase in difficulty. Data from fifty-three children (ages 8-18 years old; 22 females, 31 males) and their parent training aid are included in the following results. Parent stress was captured using the Parenting Stress index (PSI). Using a hierarchical multiple regression approach, we examined whether their child’s difficult behaviors (PSI-DC) and parental distress (PSI-PD) interact with treatment group to affect treatment adherence, specifically the average number of training session per week and average active training time per session.

**Results:** When predicting the average number of training sessions per week, treatment group had a significant moderating effect with PSI-DC scores ($B = -.04, t(49) = -2.08, p = .04$), such that children in the adaptive treatment group whose parent reported more stress related to their child’s difficult behavior trained for fewer sessions per week. This association was not observed in the non-adaptive group. The addition of parental distress did not account for a significant amount of variance in training sessions per week. When predicting the average amount of active training time per session, the interaction between treatment group and PSI-DC scores was significant ($B = -.22, t(48) = -2.19, p = .03$). Children in the adaptive treatment group whose parent reported more stress related to their child’s difficult behavior spent less time actively training. The association was reversed for the non-adaptive group. The addition of PS-PD scores did not incrementally predict a significant amount of variance beyond the PSI-DC.

**Discussion:** While parents of children with intellectual disabilities, including FXS, often experience heightened stress, less is known about the degree to which stress and mental health impacts treatment when parents are involved. In this study, results suggest that the child’s difficult behavior, but not the parent’s own experience of stress per se, impacted treatment adherence (although adherence was generally high). Future analyses to be presented will examine the impact of mental health (e.g. anxiety, depression) on treatment adherence. It may be important for investigators to account for parent, or parent-child factors that may mediate the efficacy of various treatments for children with intellectual disabilities.

**References/Citations:**

Symposium Title: Prenatal Risk Factors for Autism Spectrum Disorders: Advancing our Understanding through Integrated Epidemiological and Preclinical Studies

Chair: Melissa D. Bauman¹

Overview: Given the prevalence and societal impact of ASD spectrum disorders (ASD), there is an urgent need to identify risk factors, determine the underlying neural basis and ultimately develop novel therapeutic and pharmacological interventions. This is a daunting challenge for ASD research because of the inherent complexity of the human brain, our lack of understanding of disease neurobiology underlying ASD and the incredible heterogeneity of this disorder. There is a growing consensus that ASD is actually a number of distinct brain disorders each caused by a complex interplay of different genetic and environmental factors. Here we highlight progress in our understanding of prenatal risk factors that have been identified through epidemiological studies and then systematically evaluated in preclinical models. New findings from recent ASD epidemiological studies investigating environmental exposures, genetic factors, and their interplay will be presented. Preclinical studies evaluating in utero exposure to (i) valproic acid, (ii) autism associated maternal auto-antibodies and (iii) maternal immune activation will be highlighted as examples of basic science approaches. The discussion will focus on maximizing translational studies to advance our understanding of ASD.

Paper 1 of 4

Paper Title: Epidemiology of Autism Spectrum Disorders: Investigating Perinatal Risk Factors

Authors: Lisa Croen²

Introduction: Although the initial manifestations of autism spectrum disorders typically do not appear until several months or years after a child is born, the results of research studies conducted over the past 50 years provide overwhelming evidence that factors operating around the time of pregnancy and birth are at play.

Methods: Epidemiologic studies have played a crucial role in the identification of prenatal risk factors for autism, and in directing basic science studies focused on elucidating underlying biologic mechanisms.

Results: In this talk, findings from recent studies investigating the role of maternal immune system function during pregnancy in autism will be presented. Studies utilizing biospecimens collected from expectant mothers and their newborns will be highlighted.

Discussion: Future directions and preclinical approaches to further evaluate findings from human epidemiology studies will be discussed.

¹ University of California, Davis
² Kaiser Permanente Division of Research
Paper Title: Prenatal Valproic Acid Exposure as a Preclinical Model for the Study of Autism Spectrum Disorders (ASD): Assessing ASD-Like Social and Communicative Behaviors in Rats

Authors: Sarah Raza\textsuperscript{3}, Bryan Kolb\textsuperscript{4}, Sergio M. Pellis\textsuperscript{4}, Robbin Gibb\textsuperscript{4}

Introduction: Autism Spectrum Disorder (ASD) is characterized by impairments in social interactions, communication, and repetitive behaviors. Currently, preclinical research is focusing on the development of environmental animal models of ASD to better understand the underlying neurobiological mechanisms. However, validation of these animal models is contingent upon the ability to simulate the clinical features of ASD, such as deficits in social function. The present study assessed whether juvenile rats exposed prenatally to valproic acid (VPA) exhibit deficits in social play that are consistent with the socio-communicative deficits in ASD. Given the cognitive, emotional, and communicative demands on animals during social play, the present study provides a detailed analysis of the social play of VPA-exposed rats.

Methods: Pregnant Long-Evans dams were administered an acute oral dose of VPA (800mg/kg) on gestational day 12.5 (Raza et al., 2015). Thirty-two female rats (16 VPA, 16 control) born to the VPA-exposed and control dams were utilized. Juvenile play fighting and the associated ultrasonic vocalizations (USVs) were assessed between PN29-PN34. To investigate playful interactions, the frequency of playful attack, defensive tactics, and the rate of USV calling per bout of play fighting were examined. Non-playful behaviors - frequency of mounting and body shakes - were also assessed. It was predicted that VPA-treated rats should play less than controls and the frequency of emitting USVs would be reduced.

Results: While VPA and control rats did not differ in their frequency of launching attacks or in the probability of defending against an attack, significant differences in the use of defensive tactics were observed. That is, VPA rats were significantly less likely to turn to supine and more likely to remain standing when defending themselves, suggesting that in the VPA rats bodily contact is limited. Moreover, although the VPA rats tended to vocalize less frequently overall during play, the difference was not statistically significant. The rate of calling, however, revealed that the VPA rats called significantly less often than expected for the number of times they initiated play, suggesting reduced communicative ability. With respect to non-playful behaviors, VPA rats mounted each other more often than the control rats and were more likely to perform body shakes, indicative of altered sexual behavior and stereotypy.

Discussion: The present study supports the validity that in utero exposure to VPA produces a viable rodent model for the study of ASD, especially with respect to studying sociality. While the VPA-treated rats appeared to be as motivated to play as control rats (i.e., comparable rates of playful attack), they limited the contact involved in playful wrestling. It appears that the rewards offered by playing may have been altered, reducing the pleasure to be gained from the physical contact usually associated with play. This may also explain the reduced rate of emitting USVs relative to the amount of play initiated, reflecting a deficit in communicative ability. This discordance between vocalizing and play, and the reduction in contact-promoting tactics, may reflect altered socio-communicative function and dysregulation of pragmatics in social relations, characteristics of ASD. Furthermore, elevated frequencies of mounting and body shaking at this early developmental stage may be indicative of accelerated sexual maturation and stereotypic behavior, two additional characteristics of ASD. These data suggest that prenatal VPA exposure disrupts some aspects of being able to communicate effectively and engage partners in dynamic interactions - deficits consistent with those seen in ASD - thus, validating the VPA rodent model as a useful preclinical tool to study ASD.

\textsuperscript{3} University of Alberta
\textsuperscript{4} University of Lethbridge
**Paper Title:** Maternal Anti-Fetal Brain IgG Autoantibodies and Autism Spectrum Disorders: Translational Approaches to Determine Underlying Mechanism

**Authors:** Judy Van de Water

**Introduction:** Several studies have found a correlation between the presence of circulating maternal autoantibodies and neuronal dysfunction in the neonate. Specifically, maternal anti-brain autoantibodies, which may access the fetal compartment during gestation, have been identified as one risk factor for developing Autism Spectrum Disorder (ASD).

**Methods:** Studies by our laboratory elucidated seven neurodevelopmental proteins recognized by maternal autoantibodies, whose presence is associated with a diagnosis of maternal autoantibody related (MAR) autism in the child. Preclinical approaches utilizing mice, rat and nonhuman primate models have been carried out to evaluate the underlying neurobiology of autoantibody-mediated pathology.

**Results:** Animal model studies using gestational transfer of purified IgG from mothers of children with ASD, have shown that the autoantibodies associated with MAR autism induce long-term behavioral changes in gestationally-exposed offspring. Recent convergent findings from both mouse and nonhuman primate models indicate that prenatal exposure to these antibodies impact brain growth trajectories and parallel features from clinical ASD studies.

**Discussion:** A better understanding of the mechanisms that drive autoantibody generation and autoantibody-mediated pathology is necessary to further translate this research into a clinical application. Future preclinical research efforts will be discussed.
Paper Title: Prenatal Immune Challenges and Altered Neurodevelopment: Novel Findings and Future Directions from Preclinical Models in Rats and Nonhuman Primates

Authors: Melissa Bauman¹

Introduction: Women exposed to infection during pregnancy have an increased risk of giving birth to a child who will later develop autism, schizophrenia (SZ) or other central nervous system (CNS) disorders. In order to implement preventative prenatal care approaches, we much first understand why some, but not all, women who experience infection during pregnancy have a child who will later develop a neuropsychiatric disease. A number of factors, including genetic susceptibility, the sex of the fetus, the intensity of the infection and the maternal and/or fetal response, as well as the precise timing of the immune challenge likely influences the degree to which prenatal immune challenge alters fetal brain development and may ultimately determine which disease phenotype (SZ, autism, other CNS disorders) is expressed.

Methods: Sophisticated animal models are needed to understand how these genetic and environmental risk factors contribute to an increased risk of neuropsychiatric disorders in exposed offspring and to develop novel preventative strategies. Here we present novel findings from two preclinical models of maternal immune activation (MIA) utilizing the nonhuman primate and the laboratory rat. Pregnant dams from each species were injected with a viral mimic, polyic, to stimulate the maternal immune response. Longitudinal brain and behavioral development of the offspring were evaluated to determine the impact of prenatal immune challenge.

Results: New findings from the nonhuman primate model indicate that prenatal immune challenge yields behavioral impairments in sensorimotor gating relevant to a number of neurodevelopmental disorders. Novel findings from the rat model indicate that the severity of the maternal immune response, as indexed by cytokine profiles and sickness behaviors, influences offspring neurodevelopment.

Discussion: A better understanding of the mechanisms that drive maternal immune activation pathology is necessary to further translate this research into a clinical application. Preclinical models are critical for these efforts. The strength of integrating cross-species preclinical approaches will be discussed.
2016 Gatlinburg Conference Symposium

Symposium Title: Diagnosis and Services among Vulnerable Children from Diverse Backgrounds with ASD

Chair: Susan L. Parish (Presenter: Sandra M. Magaña)

Discussant: Leann E. Smith

Overview: The experience of a child receiving an autism spectrum disorder diagnosis and associated services varies widely based on the characteristics of the child and his or her family. This symposium will explore the experiences of children with ASD who are Latino or in the foster care system. The four studies presented highlight: (1) low rates of speech and occupational therapy use among foster care youth with ASD; (2) limited specialty care receipt among Latino children who have severe impairments; (3) Latino parents are more likely to receive a negative response from their child's health care providers when they relate their developmental concerns; and (4) validation of the ADI-R in a US-based, Spanish-speaking Latino population. Together, the papers in this symposium shed new light on the service needs and utilization of highly vulnerable children with ASD.

Paper 1 of 4

Paper Title: Prevalence and Correlates of Service Use among Children in Foster Care with Autism Spectrum Disorder

Authors: Lucy Bilaver, Judy Havlicek

Introduction: Across the United States and in Illinois, child welfare systems serve disproportionately high rates of maltreated children with emotional, behavioral, and developmental disabilities. Because the prevalence of autism spectrum disorder (ASD) in the child welfare system has exceeded the prevalence in the general population in some states, child welfare administrators need to understand the extent of available Medicaid reimbursed services. Educational interventions are considered the cornerstone of non-medical management of ASD. These interventions include behavioral strategies and habilitative therapies such as speech, physical, and occupational therapy. The purpose of this paper is to shed light on a unique population of children with ASD and describe the prevalence and correlates of their Medicaid reimbursed services (educational interventions) for the first time. We focus on Illinois foster children with ASD during FY 2013. Children in care with ASD are compared with children in care with other developmental diagnoses (DD) and children without ASD or other DD.

Methods: The study uses linked administrative data from two state agencies in Illinois. The study population includes 9,818 children between the ages of 3 and 21 who were continuously eligible for Medicaid through the child welfare system during state fiscal year (FY) 2013. Linked Medicaid claims were used to identify 459 children with ASD based on claims with ICD-9-CM diagnosis codes 299.0, 299.8, or 299.9. Another 3,161 youth had a developmental diagnosis other than ASD during FY 2013. Rates of service utilization are compared with data from of a nationally representative survey of US children with ASD, and logistic regression is used to identify demographic and child welfare related factors associated with the odds of service use.

Results: One third of children ages 3-21 with ASD in the study population received some speech therapy during the FY (33%) while only 11.3% received occupational or physical therapy. Just under half (49.5%) received some type of mental health, social skills, or behavioral modification service. When compared with nationally representative rates of service use, foster children with ASD tend to have lower rates of speech and occupational therapy, but equal or higher rates of mental health, social skills or behavioral modification services. Transition-aged youth (18-21) had some of the lowest rates of service, particularly mental

1 Brandeis University
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health, social skills, and behavioral modification (22.7%). After controlling for demographic and placement characteristics, children with ASD were 13% more likely to receive at least one of the services examined when compared with children with other DD (odds ratio, 1.13, p<0.0001). Transition-aged youth were less likely to be receiving at least one of these services than younger children after controlling for other demographic and placement characteristics.

Discussion: The low rates of speech and occupational therapy among foster youth with ASD when compared to the general population is concerning. If youth in foster care with ASD do not receive the necessary treatment to manage their condition and minimize core deficits, their prospects for independence are compromised. Child welfare systems have an incentive to attend to the specific service needs of the increasing numbers of children with ASD in order to improve outcomes for these children at discharge or during the transition to adulthood.

Paper 2 of 4

Paper Title: Severity and Latino Ethnicity in Specialty Services for Children with Autism Spectrum Disorders

Authors: Sandra Magaña5, Susan Parish1, Esther Son6

Introduction: Children with autism spectrum disorder (ASD) experience a range of severity levels characterized as levels of support they need for everyday functioning. By this definition, greater levels of severity should warrant greater use of services and supports among children with ASD. In previous studies, Latino children with ASD in the USA have been shown to have lower access to diagnosis and treatment services than White children. However, none have examined service use in relation to severity. In this study, we examined whether there are ethnic disparities between Latino and White children with ASD in specialty autism-related services, and whether condition severity moderate the relationship between ethnicity and receipt of autism services.

Methods: We used data from the Survey of Pathways to Diagnosis and Services, a supplement to the National Survey of Children with Special Health Care Needs and analyzed current use of four specialty services commonly used by children with ASD: behavioral interventions, occupational therapy, social skills training, and sensory integration. The analytic sample included non-Latino White (n=1,063) and Latino (n=120) children with any ASD. When weighted, the sample represented 431,525 White children and 85,154 Latino children with any ASD. Logistic and Linear regressions were used to determine the relationships of ethnicity and severity to the outcome service variables, adjusting for demographic variables. An interaction term of ethnicity and severity was created to test for interaction effects.

Results: We found that children whose conditions were more severe were twice as likely to receive each of the services as children who were less severe. Being Latino was significantly related to lower total service use. Furthermore, Latino children with ASD who had severe conditions received fewer specialty autism-related services than White children with similarly severe conditions. These disparities were evident despite the fact that the sample of Latino children in this data were more privileged than the general US Latino population given that it was an English speaking and relatively high income sample.

Discussion: Future research is needed to investigate factors that contribute to Latino children who have greater severity receiving fewer services than White children. Assertive policy initiatives are needed to address these disparities and ensure that these highly vulnerable children with severe conditions receive appropriate services and supports.

5 University of Illinois at Chicago
6 College of Staten Island
Paper Title: Examining the Validity of the Spanish ADI-R in a U.S.-Based Latino Population

Authors: Sandra B. Vanegas, Sandra Magaña, Miguel Morales, Ellyn McNamara

Introduction: The use of valid and reliable diagnostic tools in Autism Spectrum Disorders (ASD) is critical, as it is often the basis for a clinical diagnosis and access to services. The Autism Diagnostic Interview-Revised (ADI-R; Lord et al. 1994) is a ‘gold-standard’ parent interview that evaluates children's past and current development. Few studies have evaluated the use of translated versions of the ADI-R (e.g., Magaña & Smith, 2013). Validity studies are needed to ensure that translated versions of the ADI-R hold cultural equivalency and maintain similar psychometric properties as the English version. The current study evaluates the use of the Spanish ADI-R in a U.S.-based Spanish speaking Latino population of children with ASD and children with DD.

Methods: The current study included Spanish-speaking Latino parents of children and adolescents who were between 4 and 16 years of age and received a clinical diagnosis of ASD or a Developmental Delay (DD). The official Spanish version of the ADI-R was administered by trained interviewers. Clinical diagnoses were established by review of medical records. The final sample included parents of children with ASD (n = 29) and children with DD (n = 21).

Results: Converted discriminant scores were used as recommended by the updated lifetime algorithms (e.g., Rutter et al., 2003). Analyses of discriminant validity found that the Reciprocal Social Interaction and the Restricted, Repetitive Behavior domains were significantly different between children with ASD and children with DD. The Verbal Communication domain approached significance, however, no differences between groups were found in the Nonverbal Communication domain. Analyses of clinical validity found that the Spanish ADI-R had moderate sensitivity (69%), specificity (76.2%), and positive predictive value (80%). Inspection of individual domains and age of first concern finds that the restricted interests and repetitive behaviors domain had the highest sensitivity (79.3%), specificity (76.2%), and positive predictive value (82.1%).

Discussion: These findings suggest that the Spanish ADI-R exhibits moderate discriminant validity within the Reciprocal Social Interaction and the Restricted, Repetitive Behavior domains, however, only the Restricted and Repetitive Behavior domain demonstrated good sensitivity, specificity and positive predictive value. Overall, the Spanish ADI-R had moderate validity in use with a U.S.-based Latino population, although lower than other studies of translated versions of the ADI-R (e.g., Becker et al., 2012; Lampi et al., 2010; Tsuchiya et al., 2013). Further studies are needed to evaluate culturally sensitive tools in identifying ASD.

References/Citations:


Paper 4 of 4

Paper Title: Ethnic Disparities in Health Care Providers' Response to Parent Concerns

Authors: Susan Parish\(^1\), Sandra Magaña\(^3\), Esther Son\(^6\)

Introduction: Diagnosis of autism at a young age is critical because it allows children to begin early intervention as young as possible, and thereby maximize the benefits. Research indicates interventions are most effective in younger children, and intervention yields fewer improvements in older children (Ben Itzhak & Zachor, 2011). Conversely, as early ASD diagnosis is critical in improving children's functioning, some of the racial and ethnic disparities in severity and outcomes among children with ASD may be explained by the vast disparities in age at diagnosis among children of color (Gourdine, Baffour & Teasley, 2011). This study aimed to understand whether there were ethnic disparities in when parents noticed developmental delays in their children, and whether there were ethnic disparities in health care providers’ responsiveness to parent-reported concerns.

Methods: The results of this study are derived from an analysis of the 2011 Survey of Pathways to Diagnosis and Services (Pathways Survey), which was a subsample of families who participated in the 2009-2010 National Survey of Children with Special Health Care Needs (n=70 Latino children and 411 non-Latino white children with autism). The independent variable was whether the child's ethnicity was Latino. The dependent variables were child’s age at which the parent noticed developmental concerns, and whether the child’s health care provider responded proactively or negatively to parent-reported concerns. Multivariate logistic regressions models were estimated, controlling for a range of socio-demographic characteristics, including parent education, whether the family lived in an urban area, child's insurance type, whether the child had a usual source of care, child’s age and severity of condition.

Results: There were no significant differences in child's age when Latino and white parents were first concerned about their child’s development or in the child’s age at autism diagnosis. Latino parents were significantly more likely to report receiving a negative response from their health care provider after bringing up concerns about their child’s development. These results persisted after controlling for all model covariates.

Discussion: There were no differences in the time at which parents recognized developmental problems in their children who were later diagnosed with autism. However, even though Latino and non-Latino white parents notice their children's problems at the same time, health care providers responded differentially to Latino and non-Latino White parents. Once concerns are reported, Latino children were more likely to receive a negative response from their child’s health care providers. Health care providers and policymakers must take assertive action to eliminate ethnic disparities in the responsiveness of health care providers.

References/Citations:
Symposium Title: Experimental Analysis and Intervention for Severe Behavior Problems of Individuals with Rett Syndrome and ASD via Remote Videoconferencing

Chair: Jennifer McComas 1

Overview: Access to experts who can provide effective individualized intervention for self-injurious behavior (SIB), aggression, and communication is limited for individuals with neurodevelopmental disorders such as Rett syndrome and autism spectrum disorders (ASD). Preliminary research has demonstrated that videoconferencing can be an effective technology for providing remote consultation to families of young children with ASD. This symposium will feature work at the University of Minnesota to expand the application of videoconferencing for conducting experimental analysis and effective intervention for severe SIB, aggression, and limited formal communication in individuals with Rett syndrome and ASD. This symposium will begin with an overview of behavioral teleconsultation including its early development and current applications. The overview will also include a brief demonstration of how the technology is used and adapted to a variety of situations. Next, Brittany Pennington will present a study conducted with young girls with Rett syndrome and who have limited formal communication repertoires. Consultation was conducted remotely with parents who taught their daughters to use formal communication systems across three contexts. Stephanie Benson will present the next study in which the experimenters successfully coached a parent remotely via teleconsultation to conduct the conditions of a functional analysis and then intervention for a child with ASD who engaged in SIB. Adele Dimian will deliver the final presentation demonstrating the effects of remote coaching with group home staff to conduct an analysis and intervention for aggressive behavior of a young adult diagnosed with ASD who resided in a group home. After the data-based presentations, the chair will facilitate a discussion with the audience focused on issues relevant to expanding telehealth implementation including generalization of intervention effects and issues of implementation including fidelity and maintenance in settings such as group homes.

Paper 1 of 4

Paper Title: Applications and Extensions of Remote Videoconferencing to Conduct Experimental Analyses and Intervention for Severe Behavior Problems and Limited Communication Repertoires of Individuals with Neurodevelopmental Disorders

Authors: Jennifer McComas 1

This presentation will consist of an overview and introduction to the three data-based interventions that will follow. A brief background of the use of remote videoconferencing for the analysis and intervention of behavior problems will be followed by an overview of the current applications of remote videoconferencing. A demonstration of the technology will also be provided to provide a foundation for the three presentations that will follow.

Paper 2 of 4

Paper Title: Telehealth as A Mechanism for Coaching Parent-Implemented Communication Intervention for Children with Severe Neurodevelopmental Disabilities

Authors: Brittany Pennington 1, Jessica Simacek 1, Jennifer McComas 1

1 University of Minnesota
Introduction: Early and intensive intervention is strongly recommended as critical for children with autism spectrum disorder (ASD) and related severe neurodevelopmental disabilities such as Rett syndrome to reach optimal outcomes. However, there are numerous barriers to accessing needed intervention for this population. The purpose of the current study was to determine the effectiveness of parent-implemented communication assessment and intervention with coaching via telehealth (i.e., video conferencing) on the acquisition of early communication skills for three young children (3.5-4 years old) with severe neurodevelopmental disabilities.

Methods: Assessment included a structured descriptive assessment and functional analysis to identify idiosyncratic/potentially communicative responses and communicative contexts for each child. Functional communication training (FCT) was implemented as intervention to increase augmentative and alternative communication skills. Parents conducted all assessment and intervention sessions with remote coaching by experts in functional analysis and FCT via telehealth technology. The effects of FCT were evaluated using an adapted, multiple-probe design across three contexts for each child.

Results: Prior to intervention, none of the children engaged in reliable or easily recognizable communication forms; however, during FCT intervention all three children acquired the targeted communication skills.

Discussion: The results demonstrate the effects of functional reinforcement on communicative responses of young children with severe neurodevelopmental disorders. Implications from this study support the efficacy of parent-implemented intervention delivered with coaching via telehealth and the potential for future research to examine telehealth within a service delivery model to increase access to services for children with severe neurodevelopmental disorders and their families.

References/Citations:

Paper 3 of 4

Paper Title: Using Telehealth to Conduct Functional Analysis and Functional Communication Training for Challenging Behavior

Authors: Stephanie Benson¹, Kelsey Quest², Adele Dimian¹, Jennifer McComas¹

Introduction: Children with developmental disabilities sometimes engage in challenging behavior which if left untreated can persist over time and interfere with and individual's ability to learn (Didden et al., 2012). Functional analysis (FA) is a tool that has been demonstrated to be effective across a variety of settings and interventionists (Hanley et al, 2003). The functional reinforcer(s) identified in the FA can be differentially applied to an appropriate alternative response during functional communication training (FCT) to effectively compete with the challenging behavior. FCT has been repeatedly and reliably shown to be an effective tool in reducing challenging behavior (Carr & Durand, 1985, Iwata et al., 1994) when based on the results of an FA. There is a growing need for trained behavior analysts who can conduct these assessments in rural areas of the country. One potential way to address this need is through the use of telehealth technology. The purpose of the current study was to examine the use of telehealth for conducting FBA and FCT for a boy with Autism spectrum disorders (ASD) who engages in challenging behavior.

¹ Indiana University
Methods: A seven year old boy diagnosed with ASD was referred for this study through a local service agency in Minnesota due to his engagement in self injurious behavior. A functional analysis interview was conducted with the child’s mother over the telephone. A structured descriptive assessment (SDA) was conducted followed by a functional analysis of the challenging behavior. After the function of the behavior was identified, functional communication training (FCT) was conducted to teach the child an appropriate communicative alternative to replace the challenging behavior. All analyses and FCT were conducted in the child’s home with the parent with live coaching via telehealth.

Results: The results of the functional analysis revealed that his challenging behavior was most sensitive to restricted access to a tangible item. Results of FCT indicated that the child was successful in learning a communicative replacement for his challenging behavior.

Discussion: This study extends previous research using telehealth to conduct FA and FCT for a child with challenging behavior. These results suggest that telehealth was successful in identifying a function for the challenging behavior and the use of FCT via telehealth was successful in decreasing the challenging behavior and increasing the communicative alternative. Future research should seek to replicate these findings and include a cost analysis for delivering services in home or at a site versus using telehealth technology.

References/Citations:

Paper 4 of 4

Paper Title: Assessment and Intervention for Challenging Behavior Via Telehealth in A Group-Home Setting

Authors: Adele Dimian¹, Jessica Simacek¹, Jennifer McComas¹

Introduction: Preliminary studies have demonstrated that telehealth technology can be used to remotely coach parents to implement behavioral procedures related to the treatment of challenging behavior (i.e., tantrums, self-injury, aggression) for individuals with autism (Seuss et al., 2014; Wacker et al., 2013a; 2013b). Due to the high level of need associated with services for people with autism, and limitations due to geographic location, there is a need for access to service providers with expertise in addressing challenging behavior and intervention beyond the geographic proximity to university clinics or other centers with expertise in addressing severe challenging behavior. Group home settings in particular can have high staff turn-over and need support with addressing challenging behavior. To date, there have been no published studies conducting assessments for challenging behavior via telehealth in a group home setting. The purpose of the current study was to coach staff in a group home via telehealth to implement behavioral assessments and a reinforcement-based intervention to address aggression exhibited by an adolescent with autism spectrum disorder (ASD).
Methods: The participant was a 17-year-old male with a diagnosis of ASD. He engaged in a sequence of challenging behavior that typically started with noncompliance and verbal refusals to complete a demand or task. The noncompliance then would escalate to verbal threats (e.g., "I want to hit you"), followed by physical aggression in the form of hitting, kicking, and or scratching. A structured descriptive assessment (SDA) was implemented via telehealth (i.e., video conferencing over Google Hangout with the group home staff) with coaching from the research team. All sessions were approximately 5 min long and were conducted by the staff. A Bluetooth device was used to coach the staff through each condition over the telephone. Antecedents in the form of demands and diverted attention were observed and aggression or attempts was recorded. No programmed consequences were provided for challenging behavior and the staff were instructed to attend to the challenging behavior as they typically would. The results of the SDA were then used to create a treatment package that included shortening demands, using a token board to signal how much work the participant had to do, and differential reinforcement of alternative (DRA) behavior. The treatment package was implemented using an ABAB reversal design. Three different staff members implemented the sessions and procedural fidelity was measured with a checklist for each treatment session conducted.

Results: The SDA results showed higher rates of aggression (average 1 per min) during demand conditions compared to diverted attention and control conditions. The assessment was implemented across 3 days with three different staff members. Following the SDA, the treatment package was implemented and results indicated decreases in aggression and increases in latency to comply with demands. Data were variable across the different staff members implementing the intervention. Procedural fidelity ranged from 70% to 100% and covaried with instances of aggression.

Discussion: Overall, the treatment package reduced instances of aggression to zero levels and increased latency to comply with demands to within 2 s. Aggression was observed during the treatment conditions when procedural fidelity was below 85% and varied across staff members. These results show that telehealth may help support assessment and intervention of challenging behavior occurring in group home settings with individuals with disabilities. Ongoing training and coaching may be needed to ensure implementation fidelity over time; telehealth could help facilitate service delivery among residents within group homes, but more research is warranted.

References/Citations:
**Symposium Title:** Social Experiences of Adolescents and Adults with an Autism Spectrum Disorder: Implications for Development

**Chair:** Julie Lounds Taylor

**Overview:** In this symposium, we examine the social lives of adolescents and adults with an autism spectrum disorder (ASD). Specifically, using four different datasets, we will present data on the implications of social activities, victimization, and friendships for individual and family functioning. The first two abstracts focus on peer victimization. Bishop and colleagues report on the specific behaviors and characteristics that place youth with ASD at risk for victimization. Then, Adams and colleagues present data on the implications of peer victimization for the academic functioning of adolescents with ASD. The next abstracts examine the role of social participation and friendships. Taylor examines how social and recreational activities change during the transition to adulthood, and how that change impacts the mental health of youth with ASD. Finally, Smith and colleagues examine friendships and social participation among adults with ASD and an intellectual disability compared to those with fragile X syndrome, as well as how social activities influence family functioning. We will end with an integrated discussion about the implications of social experiences for youth and adults with ASD, with a focus on future research directions and intervention.

**Paper 1 of 4**

**Paper Title:** Individual Behavioral Characteristics Associated with Negative Peer Experiences in Adolescents with an Autism Spectrum Disorder

**Authors:** Somer Bishop, Ryan Adams

**Introduction:** Across studies, parent-reported rates of bullying among individuals with autism spectrum range from 40% to as high as 94%. This is concerning given the well-established links between bullying and poor psychological outcomes, including depression and suicidality. Unfortunately, the relationships between individual characteristics and negative peer experiences in ASD have not been carefully explored, leaving unanswered the critical question of why adolescents with ASD experience increased rates of bullying (Schroeder, Cappadocia, Bebko, Pepler, & Weiss, 2014). Information about which specific characteristics are associated with different types of bullying is needed to create more targeted interventions.

**Methods:** Data were obtained from parents enrolled in the Interactive Autism Network (IAN) who completed the Bullying and School Experiences of Children with ASD Survey (BSE; see Zablotsky, Bradshaw, Anderson, & Law, 2013). A total of 1221 parents completed the BSE survey; the current study focused only on those parents with children in 7th-11th grade who spent at least half of the school day in a mainstream setting (n=279). Participants on average were 12.8 years of age, primarily male (82.1%) and white (93.5%), and mostly attended public school (90.7%). Logistic regression was used to examine the associations between specific behaviors often exhibited by adolescents with ASD and different types of peer victimization. Gender and age were also added to each model as control variables.

**Results:** Females (odds ratio = 1.66, p<.05) and older adolescents (odds ratio = 1.12, p < .05) were generally more likely than males and younger adolescents to experience the various types of victimization. Adolescents who had frequent meltdowns (odds ratios ranged from 2.42-4.12, p<.05), rigid rule keeping (odds ratios ranged from 1.34-2.46, p<.05), and/or poor hygiene (odds ratios ranged from 2.52-2.79, p<.05) were more likely to experience verbal bullying, being ignored, and being provoked. Self-injurious behaviors were also associated with higher rates of being ignored and being provoked (odds ratios ranged from 1.74-
2.25, p<.05). Physical victimization was only more common among those reported to have frequent meltdowns. (odds ratio = 1.94, p<.05). Interestingly, adolescents with repetitive behaviors and/or verbal tics were reportedly less likely to be verbally victimized (odds ratios ranged from .34-.54, p<.05).

**Discussion:** These findings provide preliminary evidence that specific behaviors exhibited by some adolescents with ASD may put them at particularly high risk for experiencing different types of peer victimization. While anti-bullying interventions should of course seek to modify the behavior of bullies, these interventions might also be improved by targeting the behaviors of adolescents with ASD. In particular, focusing on reducing behaviors, such as those identified in the current study, that are empirically shown to be linked to peer victimization, may serve as a potential means of decreasing negative peer experiences for adolescents with ASD.

**References/Citations:**

**Paper 2 of 4**

**Paper Title:** Testing Associations between Peer Victimization and Academic Problems in Adolescents with an Autism Spectrum Disorder Who Spend a Majority of Their School Day in a Mainstream Setting

**Authors:** Ryan Adams³, Amie Duncan³, Somer Bishop²

**Introduction:** Over half of all students with ASD spend a significant amount of the school day in a general education classroom setting. Unfortunately, many students with ASD in mainstream settings exhibit gaps between their intellectual ability and academic achievement and experience high rates of other negative academic outcomes such as disobedient behavior at school and fearing school. Overall, these studies indicate a critical need to understand more about why so many adolescents with ASD experience poor academic outcomes in inclusive settings. Many studies implicate social functioning deficits as a likely contributor to academic problems, which is significant since these adolescents are often targets of peer victimization. Rates of peer victimization among individuals with ASD are significantly higher than in typically developing individuals and in those with other disabilities. The current study examined the associations between multiple forms of peer victimization and multiple types of academic outcomes to identify specific types of peer victimization experiences to target towards improving academic outcome of this group.

**Methods:** S4 adolescents (M = 14.62 years, SD= 2.25) and one of their parents completed a one-hour study visit in a hospital clinic space. All met inclusion criteria: diagnosed with ASD by a medical professional using a comprehensive battery of surveys and interviews, between the ages of 10-17, had fluent language, and spent a majority of their day in a mainstream setting at school. To measure peer victimization, adolescents reported on twelve items measuring four subscales: verbal, relational, physical, and social victimization especially salient for those with ASD (e.g., How often do other kids make fun of or tease you when you try to talk to them?). All scales had acceptable internal reliability (all Cronbach's alphas ranged from .89 -.78) except physical (Cronbach's alpha = .55). School based well-being as measured by three items completed by the parent on the Child Behavior Checklist (CBCL): disobedient at school, fears school, and poor school work.
Results: A series of multiple regressions were performed. Each regression used one of the three academic measures as an outcome and a used one of the four forms of victimization as a predictor while controlling for age. For parent reports of being disobedient at school, adolescent reports of verbal (R square change = .33), relational (R square change = .15), and ASD related (R square change = .40) victimization were associated, but not physical. For parent reports of fearing school, only verbal victimization (R square change = .08) was associated. Finally for parent reports of poor school work, verbal (R square change = .12) and ASD-related (R square change = .13) peer victimization were associated.

Discussion: Overall, the findings showed that verbal victimization was consistently associated with poor academic outcomes, as reported by parents. ASD-related victimization also had a significant impact on academic outcomes. These associations are especially interesting because they involve self-reports of victimization being linked to parent reports of academic outcomes, thus eliminating concerns of shared method variance. During the symposium, we will further explain the implications of these findings in terms of identifying those at-risk for poor academic outcomes in mainstream settings and designing interventions to address these issues.

References/Citations:

Paper 3 of 4

Paper Title: Changes in Social Activities for Youth with an Autism Spectrum Disorder during the Transition to Adulthood

Authors: Julie Lounds Taylor

Introduction: Although low rates of social participation are commonly reported for youth with autism spectrum disorders (ASD; Tobin, Drager, & Richardson, 2014), it is not known how leaving high school impacts their social/recreational activities, nor whether changes in social activities have mental health implications. The present study uses a longitudinal design to examine changes in social/recreational participation for youth with ASD from before to after high school exit. We also examined whether changes in activities were related to internalizing symptoms.

Methods: Participants were 33 youth with ASD and their families. Data were collected at two time points: when youth were in their last year of high school, and 6-12 months after high school exit. Youth averaged 18.8 years of age at the start of the study, with a range from 17 to 22. Most (84.8%) were male and the majority was white non-Hispanic (90.9%). Just over 30% had an intellectual disability. Parental respondents included 29 mothers and 4 fathers, who were well-resourced on average (although 25% had incomes < $40,000).

At both time points, parents reported how often their son/daughter participated in 10 different activities (from 0 = less than yearly/never to 4 = several times a week). Activities were grouped into the following areas: unstructured social activities (social time with relatives, friends/neighbors, or with people from school/work); structured social activities (religious services, social events at church, formal/informal recreational activities, playing sports with others); and other activities (working on hobbies,
travel, and exercise). Average participation and a count of the number of activities were calculated for each area. Internalizing symptoms were measured both times using Achenbach’s Adult Behavior Checklist.

**Results:** The most common activity was participating in a formal or informal recreational activity (e.g., bowling, movies) and the least common was playing sports with others. Most youth spent some unstructured social time with classmates, co-workers, friends, or neighbors, but few did this regularly. For example, when in high school, 55% of youth spent some time with people from school/work outside of school/work hours, but only 15% did this once a week or more.

Youth in their last year of high school, on average, participated (to some extent) in 7.7 activities; the average decreased to 7.0 activities after high school exit, $t(32)=2.8$, $p<.01$. There were no differences in the amount of unstructured social activities or "other," primarily non-social activities, $ts(32)=.06$ and .24, respective, $p=ns$. However, there was a significant decline in structured social activities after high school - both in the average amount of participation and in the number of activities, $ts(32)=3.10$ and 2.97, respectively, $ps<.01$. The extent of decline in structured social activities was marginally related to internalizing problems when youth were out of high school, Spearman $\rho=-.33$, $p=.07$. Youth who had a greater drop in structured social activities were more likely to have borderline or clinical-level internalizing symptoms.

**Discussion:** In addition to difficulties finding vocational/educational activities (Taylor & Seltzer, 2011), these findings suggest that many youth with ASD lose some structured social activities after leaving high school. Importantly, this loss of activities might be related to internalizing problems. Further analyses will continue to examine the relations between changes in social and recreational activities during the transition to adulthood and mental health.

**References/Citations:**

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**Paper 4 of 4**

**Paper Title:** Friendships and Social Participation as Markers of Quality Of Life of Adolescents and Adults with Autism and Fragile X Syndrome

**Authors:** Leann E. Smith, Marsha R. Mailick, Jan S. Greenberg

**Introduction:** Although friendships and social participation are key domains of quality of life (QOL), little is known about these QOL outcomes for individuals with intellectual disability (ID) later in the life course and if QOL may vary by age. The present study examined the friendships, social and recreational activities, and family social networks in two distinct diagnostic groups of adolescents and adults with ID: individuals diagnosed with autistic disorder ($n=226$) and those diagnosed with fragile X syndrome (FXS; $n=81$). We explored differences between diagnostic groups as well as differences by age group (adolescent vs adult).

**Methods:** Individuals with ID were drawn from two linked longitudinal studies, one focused on families of adolescents and adults with autism and the other focused on families of adolescents and adults with FXS. Mothers reported on the adolescent or adult child’s friendships, social and recreational activities, and social impact on the family. Friendships were measured using the current rating of friendship from the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994). Data were recoded so that scores of "0" indicated "having a mutual friendship" and codes of "1" as "not having a mutual friendship". Participation in social

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and recreational activities was measured using a modified version of questions from the National Survey of Families and Households (Bumpass & Sweet, 1987). Items included spending time with friends, attending religious services, doing hobbies, and exercise. We recoded scores to approximate the number of times per year that the individual engaged in each activity in order to aid in interpretability. A total score for participation in social and recreational activities was calculated by summing all items, with higher scores indicating more frequent participation. Total negative social impact on the family was measured using items drawn from the Zarit Burden Interview (Zarit, Reever, & Bach-Peterson, 1980). Family social isolation was measured by to the question "To what extent is your family socially isolated because of your son/daughter?" on a 4 point scale ranging from "not at all isolated" to "extremely isolated."

**Results:** QOL in friendships was notably low for both diagnostic groups, with less than 20% of individuals with FXS and less than 10% of individuals with autism having any mutual friendship. Individuals with autism had fewer friendships than individuals with FXS (F=8.84, p<.01); there were no significant age or age X group effects. Individuals with autism also experienced less social and recreational participation (F=13.16, p<.001) than individuals with FXS. There was a marginally significant age by diagnostic group effect (F=3.52, p<.10), such that participation was lower for adults with autism (M=349.28) compared to adolescents (M=404.75), but participation was higher for adults with FXS (M=542.17) compared to adolescents (M=470.14). There also was a greater negative social impact on the family (F=9.46, p<.01) and greater family social isolation (F=18.69, p<.001) for individuals with autism compared to individuals with fragile X. Negative social impact and social isolation were higher for families of adolescents compared to families of adults for both diagnostic groups.

**Discussion:** Our findings indicated that, compared to individuals with FXS, not only did individuals with autism have very few reciprocal friendships and significantly reduced social and recreational activities, but they also had mothers who reported the highest levels of social isolation. Findings also suggested that these differences may be more pronounced at older ages. The lack of friendships and social activities, particularly for older individuals with autism, is a cause for concern and indicate a call for future work in understanding how to promote a high QOL for all individuals with disabilities.
Symposium Title: Training Usual Care, Community Providers to Deliver Evidence-Based Practices to Children with Autism Spectrum Disorder

Chair: Amy Drahota 1,2

Overview: The Centers for Disease Control (CDC) estimates that 1 in 68 children have autism spectrum disorder (ASD) and treatment of core symptoms of the disorder as well as co-occurring psychiatric disorders and behavioral health challenges is estimated at $268 billion annually (Autism and Developmental Disabilities Monitoring Network, 2014; Leigh & Du, 2015). Individuals with ASD receive services within a variety of service systems (e.g., education, early intervention, mental health, for-profit community-based organizations). Providers within these systems have various educational and training backgrounds, which likely impacts the implementation of evidence-based practices (EBP) and quality of services. The prevalence of ASD is increasing and, the need for providers to deliver EBP is high. Thus, research on training practices across and within each service system is critical to meet this growing public health need.

Presenters in this symposium will share data from four studies examining training practices in diverse community service settings. These studies range from describing usual training practices to evaluating the effectiveness of training protocols designed for specific contexts and providers. These studies represent the growing body of training and services research related to ASD that may have specificity to best fit within specific service contexts as well as general themes related to evidence-based training strategies. Findings have the potential to make a significant contribution to the broader services research field.

Dr. Drahota will describe a mixed methods study evaluating training-as-usual practices common among community-based organizations providing specialty intervention services to individuals with ASD (ASD-CBO); and provide information about the current status of provider training to deliver EBP to children with ASD within behavioral and mental health settings.

Dr. Reith will describe a study examining multi-disciplinary therapists’ perceptions of training modality and fidelity of implementing in a blended behavioral and developmental intervention adapted for use with toddlers in community service systems (i.e., school district early intervention program, autism-specific behavioral service agency, speech and language therapy center, hospital-based developmental center, and children’s hospital based autism clinic).

Dr. Suhrheinrich will describe a research-based training protocol for classroom pivotal response teaching (CPRT) designed for teachers and paraprofessionals and present findings from a study examining the preliminary teacher, paraprofessional, and student outcomes from a large-scale community effectiveness trial.

Dr. Chlebowski will describe a study evaluating the impact of providing a training to community mental health providers of a mental health intervention designed for delivery in outpatient and school-based mental health services on use of session organizational strategies and active teaching strategies.

Paper 1 of 4

Paper Title: Training as Usual for Community Providers Providing Specialized Services to Youth with Autism: A Lot of Methods but Little Consistency

Authors: Amy Drahota 1,2, Alexandra Smith 1,2, Lauren Brookman-Frazee 1,2

1 San Diego State University
2 Child & Adolescent Services Research Center (CASRC)
Introduction: Greater prevalence of ASD and insurance coverage for specialized autism services has increased the need for community-based organizations providing evidence-based practices (EBP) (National Conference of State Legislators, 2012). Recent systematic reviews of the efficacy and effectiveness of ASD interventions indicate that a number of established EBP are available for children with ASD (National Autism Center, 2009, 2015; Wong et al., 2015). There is recognition that implementing EBP into community settings is a challenging yet essential step for improving quality of care and access to effective behavioral health services for individuals with ASD. The NIMH Strategic Plan and the IACC identify optimizing the delivery of EBP in community settings through the use of implementation science methods as a particularly potent area of research to "bridge the gap" between EBP and community services.

Few studies have evaluated the usual care practices, training, or implementation of EBP by for-profit community-based organizations providing specialized services to children with ASD (ASD-CBO). Although ASD-CBO are contracted to provide ABA services, it is not known the extent to which services resemble those in ABA research. Further, BCBA certification requires training and supervision of non-certified staff, but little is known about how staff are trained. The purpose of this study was to evaluate training-as-usual within ASD-CBO.

Methods: A sequential mixed methods (QUAN→QUAL) design was used with quantitative and qualitative data given equal weight for the purposes of complementarity ("using each set of methods to answer a related question for purposes of evaluation or elaboration") (Palinkas et al., 2011, p. 46). This study was conducted as part of a larger partnered research study evaluating usual care practices of ASD-CBO, organizational readiness for change, provider attitudes toward EBPs, and factors impacting adoption and implementation of EBP.

Participants included 20 agency leaders (AL) from ASD-CBO and 27 direct providers (DP) in San Diego and Orange Counties. Participants responded to a survey that included questions about the availability of training at their ASD-CBO, training content and format and perceived utility. Frequency counts were conducted for quantitative data and a content analysis analyzed written narrative from the quantitative surveys. A subset of 10 AL participated in a follow up interview coded using a coding, comparison, and consensus approach (Willms, 1990).

Results: Most of the DP (96.7%) reported being provided training at their ASD-CBO. Training most commonly was "on-the-job" training (90%), occurring during supervision (80%) or in consultation with peers (73.3%). Over half DP reported regularly scheduled trainings. Training was primarily conducted by staff supervisors (80%), agency director/leaders (76.7%) or colleagues (43.3%). Overall, obtaining practical information about the application of intervention strategies was most useful to DP. Specifically, the most common description of useful training strategies included, "watching videos" (n=8), "discussions of actual use" (n=3) and "practicing with feedback" (n=3).

Primary themes from the qualitative analyses included: AL evaluating new staff to determine their training needs and various new hire training methods (e.g., co-treating until staff reach fidelity; didactics, seminars or online training; shadowing; and observation). Ongoing training involved didactics, seminars or online training for staff. Forty percent of the agency leaders felt that their ASD-CBO did a good or better job training staff than other agencies while 30% felt that their agency’s training process was not ideal.

Discussion: Training-as-usual seems to involve many different methods, depending on the timing of the training (new hire vs. ongoing) and may not include evidence-based teaching strategies derived from adult learning literature. Adequate staff training is critically necessary to increase delivery of EBP and improve quality of care; therefore, greater research to develop feasible and effective training strategies in ASD-CBO is necessary.
Paper 2 of 4

Paper Title: Training Multi-Disciplinary Early Intervention Providers in an Evidence-Based Practice for Toddlers At-Risk For ASD

Authors: Srah Rieth\textsuperscript{1\,2}, Aubyn C. Stahmer\textsuperscript{2\,3}, Lauren Brookman-Frazee\textsuperscript{1\,2}

Introduction: Increasing numbers of children with autism or risk for autism are being identified at early ages in community settings. Positive results for autism-specific interventions for toddlers have recently been published in the scientific literature, but have not yet been translated into usual care. Community providers supporting young children with ASD may not have experience with infants and/or toddlers, and may be implementing interventions that are not developmentally appropriate or do not fit the providers' theoretical background and experience. To better understand the realities of training multi-disciplinary community providers to support toddlers and their families, the current study examines therapists' fidelity of implementation in a blended behavioral and developmental intervention adapted specifically for use with toddlers in community service systems following intensive training from the model developer.

Methods: Participants included 10 therapists from 5 community agencies. Participating agencies included a school district early intervention program, an autism-specific behavioral service agency, a speech and language therapy center, a hospital-based developmental center, and a children's hospital based autism clinic. Participating therapists had at least two years of experience with infants/toddlers with autism, held an MA degree or equivalent in their discipline, and had worked at their current agency for at least one year. Therapists' self-reported theoretical backgrounds included applied behavior analysis, developmental/relationship-based approaches, and general early intervention (non-autism specific).

All participating therapists received intensive, two-day didactic training in the blended developmental and behavioral intervention approach described in Teaching Social Communication for Children with Autism (Ingersoll & Dvortskay, 2010). After the workshop, therapists practiced the intervention at their sites for one month while receiving feedback from a local intervention expert. Following the practice period, video probes were collected to assess therapists' fidelity of implementation in the intervention, and to examine areas of strength and weaknesses. After training was complete, therapists participated in focus groups, surveys and exit interviews to discuss their opinions on the training process and the intervention itself.

Results: Evaluations from therapists indicated that they felt the training prepared them to use the intervention with families and that lecture and videos were the most helpful modalities of training. Fidelity data following training indicate some areas of strength and weakness across all therapists, and some areas of strength and weakness that vary by therapists' theoretical background. As expected, strategies that matched the therapists' backgrounds were easiest for them to implement. For example, therapists with backgrounds in applied behavior analysis excelled at intervention components related to prompting for more complex responses and adjusting the prompts, but had more difficulty with modeling and expanding developmentally appropriate play skills. Data from therapist exit interviews and follow-up focus groups indicate a need for the training content to be delivered over a longer period of time, and with opportunities for hands-on practice throughout the learning period.

Discussion: Differential learning of therapists from various theoretical backgrounds suggests that training could be targeted based on prior knowledge. Therapists may particularly benefit from receiving feedback from a colleague with a differing background to promote cooperative training and facilitate learning. Data from the current study were used to inform an improved training model, both in terms of the delivery of information and the content covered. Details of the structure and topics of the improved training will be discussed.

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Paper 3 of 4

**Paper Title:** Training Classroom Teachers in an EBP for ASD: Examining Outcomes and Predictors of Skill Mastery

**Authors:** Jessica Suhrheinrich\(^1\), Aubyn C. Stahmer\(^2\), Sarah Reith\(^1\)

Classroom Pivotal Response Teaching (CPRT) is a behavioral ASD intervention adapted from Pivotal Response Training through an iterative process in collaboration with teachers and school administrators. This study provides preliminary teacher, paraprofessional, and student outcomes from a large-scale community effectiveness trial of CPRT involving 17 school districts and over 100 classroom units. The randomized waitlist control design involved three training cohorts across four academic years, with baseline, treatment and follow-up data collected. Adult learning literature and best practices in community-based training were reviewed to develop a comprehensive training protocol. Training procedures included 12 hours of small group didactic instruction, including goal setting, supervised exercises and role-playing. Individual coaching followed at weekly, then monthly intervals upon completion of didactic training, with an average of 7.6 coaching sessions overall. During coaching, teachers worked with their own students during regular classroom activities. Demographics and classroom quality ratings were collected for all teachers. Demographic, autism severity (ADOS-II), cognitive functioning (DAS or MSEL) and goal progress data were collected for all students.

Data representing 106 teachers and 270 students have been collected. Students were in preschool through 6th grade, with an average age of 5.8 years at intake. Student data demonstrate that 99% of participating students met criteria for autism, 18% scored within one standard deviation of the mean on cognitive assessments (80% below; 2% above). Participating students were 86% male and representative of the general population in San Diego County, with just under 50% of students Hispanic, based on parent report. Participating teachers were 94% female, had an average of 9.2 (r = 1-34) years teaching children with ASD and reported high levels of job satisfaction (3.64 on 1-5 rating scale) and moderate levels job related stress (2.22 on a 1-5 scale). Classroom quality data was organized by domain area (Teaming, Classroom structure, Classroom environment, Curriculum and instruction, Social/peer relationships, Challenging behaviors, Instructional climate). These variables were analyzed in relationship to fidelity of implementation (FI) of CPRT. Preliminary data indicate 75% of participating teachers met criteria for FI at least once during the study period. On average, after training, teachers reported using CPRT for 50 min/day, four days a week. Time to acquisition of CPRT and fluency of use varied by participant; analysis of relationships between teacher and classroom factors and FI of CPRT are on-going and will be discussed. A comprehensive description of teacher, paraprofessional and child characteristics will also be provided.

Results indicate CPRT is feasible in classroom settings and classroom teachers can be trained. These findings indicate collaborative adaptation of evidence-based practices may facilitate FI in community settings.

Paper 4 of 4

**Paper Title:** Training Mental Health Providers to Deliver an Individualized Mental Health Intervention for Autism Spectrum Disorders (ASD): Impact on Provider Use of Evidence-Based Intervention Strategies

**Authors:** Colby Chlebowski\(^1\), Margaret Dyson\(^2\), Lauren Brookman-Frazee\(^1\)

**Introduction:** Publicly-funded community and school-based mental health (MH) programs play an important role in caring for school-age children with autism spectrum disorders (ASD). Previous research indicates that many MH providers have limited ASD training and do not deliver strategies consistent with evidence-based (EB) practice for ASD in usual care. AIM HI (“An Individualized Mental Health Intervention for ASD”) is a clinical intervention and corresponding training model designed to address this need. AIM HI is a package of EB strategies designed to be delivered by community MH providers serving children
ages 5-13 with ASD. The AIM HI training model consists of a training workshop and six months of consultation. Examining the impact of providing training to MH providers is essential to evaluate efforts to implement evidence based practices in the community.

**Objective:** This presentation will evaluate the impact of providing AIM HI training to community MH providers serving children with ASD using data from multiple informants. Data for this study were collected as part of an ongoing, large-scale community effectiveness trial of AIM HI.

**Methods:** Participants include 142 MH providers participating in an ongoing community effectiveness trial. Multiple methods and informants were used to measure provider behavior and delivery of EB strategies in treatment, including therapist self-report, caregiver report of provider behaviors, and observational data (based on videos of therapy sessions coded by blind coders). The EB strategies examined include use of session organizational strategies (use of materials, visuals, session schedule) and Active Teaching strategies (use of psychoeducation, modeling, behavioral skill rehearsal with feedback, and positive reinforcement) to target child and caregiver skill building.

**Results:** Results highlight group differences between providers enrolled in training and those who did not receive training in regards to use of EB strategies including both session organizational strategies and Active Teaching approaches.

Trained therapists were found to implement significantly more intensive use of organizational strategies during sessions with child clients based on both therapist self-report and caregiver report. In sessions with caregivers, trained therapists self-reported using more intensive organizational strategies (F (1, 140) = 72.15, p < .001) and caregivers responses reflect a similar pattern with a trend to significance (F (1, 119) = 3.07, p = .08).

Both therapists and caregivers reported more frequent and/or intensive use of Active Teaching strategies directed to caregivers by trained providers as compared to providers who did not receive training. Caregivers also identified more frequent and/or intensive use of Active Teaching strategies by trained providers with child clients (F (1, 132) = 14.25, p < .001).

Ratings of EB strategy use by observers blind to training status using a six point scale found significantly higher use of Active Teaching directed to both caregivers (mean of 3.36 vs. 1.76; F (1, 95) = 32.19, p < .001) and children (mean of 3.51 vs. 2.70; F (1, 113) = 20.49, p < .001) by trained providers. Additionally, blind observers rated trained therapists’ intervention strategies to be more effective than strategies used by untrained therapists (mean of 4.12 vs. 2.61; F (1, 123) = 72.31, p < .001).

**Discussion:** Preliminary data indicate that provider participation in AIM HI training results in changes in MH providers' behavior and increased use of EB intervention strategies based on observer, therapist, and caregiver report. Future work will continue to expand on these findings and explore implementation and training processes to refine training strategies and increase our understanding of the process of implementing evidence based practices to providers serving children with ASD.
**Symposium Title:** Self-Injury in Developmental Disorders: Models & Mechanisms

**Chair:** Caroline Richards\(^1\), Frank Symons\(^2\)

**Discussant:** William MacLean\(^3\)

**Overview:** Tremendous advances have been made in the past 4 decades in our understanding of self-injurious behavior (SIB) among individuals with intellectual and related developmental disabilities (IDD). Despite the ‘wins’, there are myriad complexities making SIB a deeply disturbing clinical challenge and continuing scientific puzzle. Our understanding of the pathway or pathways to its development are relatively limited and remain more theoretical than empirical. We have little direct evidence that the incidence of the disorder has been affected. Nor is it clear if we can predict with enough certainty who is more or less ‘at risk’ among vulnerable populations to guide early intervention or prevention efforts. Teasing apart behavioral mechanisms has been the cornerstone of our scientific understanding of maintenance, but cases and subgroups with automatically maintained SIB present considerable clinical challenges. The question of syndrome-specific SIB and risk factors remains important as we continue to resolve and refine genotype-phenotype relations that may provide insight into specific vs. shared features of SIB comorbid with genetic ID syndromes. For all of the issues, the evidence is ambiguous and still relatively ‘thin’. We have some empirical sense of risk factors but need more depth informed, perhaps, from other models of developmental risk. We have the tools to identify automatically reinforced SIB subgroups but that translation into efficacious treatment is still empiric. The question of vulnerability is slowly surfacing but the pace of our understanding could perhaps be accelerated by committing to different approaches. To foster the discussion and cross-polinate possible approaches, we have adopted a data-blitz style symposium featuring emerging ideas with high relevance for models and mechanisms of SIB. The datablitz presentation approach provides for a unique opportunity to highlight the diversity of active SIB research programs that may lead to increased opportunities for interdisciplinary collaboration and translational research in IDD. Each presentation is designed to highlight the key underlying concept and approach and the novel data resulting from their combination.

**Paper 1 of 8**

**Paper Title:** Astroglial Activation In Macaques Exhibiting Self-Injurious Behavior: Molecular Mechanisms

**Authors:** Olivia M. Purcell\(^4\), Kim M. Lee\(^5\), Elizabeth C. Martin\(^4\), Kevin C. Chiu\(^4\), Yoojin R. Rhee\(^5\), Cagla Akay Espinoza\(^5\), Kate C. Baker\(^4\), Kelly Jordan-Sciutto\(^5\), Andrew G. MacLean\(^4\)

**Introduction:** Self-injurious behavior (SIB) can be classified as intentional, direct injuring of body tissue either with or without suicidal intent. In its non-suicidal form it is commonly seen as a clinical sign of borderline personality disorder, autism, PTSD, depression, and anxiety affecting a wide range of ages and conditions. In rhesus macaques SIB is most commonly manifested through hair plucking, self-biting, self-hitting, and head banging. SIB in the form of self-biting is observed in approximately 5-15% of individually housed monkeys. Recently, glial cells are becoming recognized as key players in regulating behaviors. The goal of this study was to determine the role of astrocytes in macaques that had displayed SIB.

**Methods:** The clinical records system at TNPRC was searched for animals exhibiting SIB, but without pharmacological intervention. Formalin fixed samples of frontal lobe (including ACC) were recovered from the pathology archives, and sectioned

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\(^5\) University of Pennsylvania
at 6μm. Quantitative immunofluorescence for GFAP, TLR2, vimentin, nestin, and E2F1 was performed as standard. Morphometric criteria were measured using Neurolucida, and proportions of activated astrocytes were determined. RNASeq analyses were performed to determine the underlying molecular basis for the changes. Age-matched animals were used as controls.

**Results:** The SIB animals exhibited shorter and less complex astrocytes that were more likely to be activated with regards to TLR2 (1) and vimentin expression. However, there was no increase in proportion of GFAP / nestin double positive cells. This was coupled with a significant decrease in E2F1 expression in both astrocytes and neurons. At the molecular level, there was increased expression of genes linked to wnt and Notch pathways.

**Discussion:** This study highlights general molecular changes in frontal brain tissues of primates, and more specific activation of astrocytes. That vimentin, GFAP and TLR2 expression were altered, but not nestin, indicates a specificity to the astrocyte activation. Molecular analyses of differentially-regulated genes were linked to neuronal differentiation, innate immune activation and vascular remodeling. Further investigation is needed to determine precisely which cells are activated, and if treatment with antidepressants reverse all the components of the activation observed (2).

**References/Citations:**

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**Paper 2 of 6**

**Paper Title:** Proopiomelanocortin (POMC) Sequencing and Developmental Delay: Preliminary Evidence for a SNP in the 3' UTR region of the POMC gene - Possible Relevance for Biological Risk and Self-Injurious Behavior

**Authors:** John A. Damerow\(^6\), Raymond C. Tervo\(^7\), Frank J. Symons\(^6\)

**Introduction:** The proopiomelanocortin (POMC) molecule has been implicated in models of self-injurious behavior (SIB) in developmental disorders, but never specifically sequenced in search of base specific polymorphisms. To date, there has only been one investigation of the POMC gene in an I/DD sample with relevance to SIB. Almost two decades ago, Sandman et al. 1 published a paper in which the POMC gene was examined from seven individuals- two participants were young brothers with autism and the other five were adult participants each with SIB and autism or autistic features. The specific genetic sequences of the samples were not investigated by Sandman et al. The goal, therefore, of this preliminary study was to conduct specific sequence analysis of all exons, and UTR sequence of POMC in a high risk sample (children with global developmental delay) with and without SIB.

**Methods:** Following IRB approval and informed consent, a clinical convenience sample was formed. The empirical focus of this preliminary study was to sequence the POMC gene in eleven children (mean age = 41.8 mo, range = 12 mo-60 mo; 73% male) with clinical concerns regarding global developmental delay, five with reported self-injury. Genomic DNA was extracted from blood samples and the POMC gene was amplified by specific oligonucleotide primers via Polymerase Chain Reaction (PCR). The

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amplified gene products were sequenced by the University of Minnesota Genomic Center and the results were analyzed using Sequencher software.

**Results:** A Single Nucleotide Polymorphism (SNP), 1130 C>T, was found in the 3' UTR region of two samples (one of whom had SIB). The program TargetScanHuman was used to predict the function of this mutation. Variant c.1130 C>T was predicted to be located in the target site of two miRNAs (hsa-mir-3715 and hsa-mir-1909) and the variant allele T may result in an increased MFE for the two miRNAs. Of the eleven samples, seven showed the same base, C, as the reference sequence. Two of the eleven samples were heterozygous for this SNP, and the final two samples showed a different base, T. One of the adult control samples had the same base at this position but the other control sample was heterozygous at this location.

**Discussion:** Further work with larger samples is needed to continue the investigation of POMC's possible function as a risk factor for the development of SIB in children with developmental delay/disability. The findings presented in this study show that the SNP found in the 3' UTR could alter the binding of miRNAs to POMC 3'UTR, thus, increasing POMC expression and affecting several biological processes with high relevance to the biology of SIB.

**References/Citations:**

Supported, in part, by NICHD Grant No. 44763, 47201.

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**Paper 3 of 6**

**Paper Title:** Environmental Impoverishment: A Rodent Model of Pathological Behavior in Autism

**Authors:** Darragh P. Devine

**Introduction:** Autism is a neurodevelopmental disorder in which deficits in social-emotional reciprocity, inadequacies in nonverbal communication, and difficulties in establishing and understanding social relationships severely limit social interactions. Engagement with the environment may be further impaired by motor stereotypes, cognitive inflexibility, restricted interests, and sensory processing disorders. Taken all together, these early difficulties may be viewed as a form of environmental deprivation. This raises the possibility that the core characteristics of autism may promote further pathological changes during development. Indeed, it has been reported that ostensibly normal children who are raised in severely impoverished environments (such as the Romanian orphans of the 1980s and early 1990s) exhibit a variety of autistic-like features, including repetitive and self-injurious behaviors [1]. These unfortunate orphans also exhibited neuropathological findings, including abnormalities in frontal cortices, basal ganglia, and limbic structures [2,3]. Although the data from studies of children with autism are not as clear, similar or overlapping neuroanatomical abnormalities have been described [4]. Accordingly, we have proposed that the early failures in social interaction and environmental engagement that characterize children with autism, cause deficits in stimulus-driven pruning of connections between frontal cortex, basal ganglia, and limbic structures [5]. We further propose that these structural and functional anomalies may be associated with aberrant repetitive and self-injurious behaviors that are frequently expressed by children with autism.

**Methods:** We are examining the behavioral and neurobiological consequences of early environmental deprivation in a rodent model. Weanling rats were placed into enriched or impoverished environments and maintained in those environments for up to 60 days. The rats in enriched environments were housed with social peers and complex stimuli (enriched condition). Some of

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the rats in impoverished environments were housed in isolation (impoverished-isolated), and some of the rats in impoverishment were housed in pairs (impoverished-social). In the final week of exposure to the enriched or impoverished environments, some of the rats were injected daily with pemoline, and the expression of self-injurious behavior was evaluated. Other rats did not receive pharmacological interventions, and connectivity between the frontal cortex and striatum is being assessed with a di-alkyl indocarbocyanine tracer (DiI).

Results: Rats in all three of the housing conditions exhibited self-injurious behaviours when treated with pemoline. However, the rats that were raised in impoverished-isolated conditions were particularly vulnerable, and they exhibited the most severe self-injury. The rats in the enriched condition were relatively resistant, exhibiting the least self-injury. Rats in the impoverishedsocial condition exhibited an intermediate amount of self-injury.

Discussion: The findings of this study demonstrate that environmental deprivation promotes vulnerability for self-injurious behavior, and that social isolation is a particularly important component of the environmental conditions that confer this vulnerability. This has implications for our understanding of the co-morbid behavioural characteristics that are often expressed by children with autism. The neuroanatomical characterizations in this study require long post mortem incubation times. Hence, we are continuing to examine the brains of the rats to determine potential structural and functional consequences of social and environmental deprivation during early development.

References/Citations:

Supported in part by a grant from the Dept of Defense - CDMRP (AR093546)

Paper 4 of 6


Authors: Adele Dimian², Jason Wolff³, Jed Elison⁴, Joseph Piven⁶

Introduction: Lifetime prevalence of self-injurious behavior (SIB) is approximately 50% among individuals with autism spectrum disorder (ASD). Once entrenched, SIB can be difficult and costly to treat. Much of the extant literature concerning possible risk factors for SIB are based on studies of adults with intellectual disabilities. Commonly identified risk factors include a diagnosis of ASD, lower intellectual functioning, communication impairment, and presence of stereotypy/repetitive behaviors (Baghdadli et al., 2003; McCintock et al., 2003; Oliver & Richards, 2015). However, there has been little empirical research utilizing a prospective research design to identify risk factors for SIB during the first years of life. The purpose of the present study was to investigate characteristics at 12 months of age predicting SIB at 24 months of age among children at high risk for ASD.

Methods: Participants were from the Infant Brain Imaging Study (IBIS), a longitudinal study of infants at familial risk for ASD. The present sample included high-risk infants with complete cognitive and behavioral assessment batteries, including Mullen Scales of Early Learning, Vineland Adaptive Behavior Scales-II, and the Repetitive Behavior Scales-Revised (RBS-R) at both 12 and 24

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months of age (n = 237). The study sample was 62.9% male. Mean ages at 12 and 24 month assessment dates were 12.54 (SD = 0.62) and 24.82 (SD =1.47), respectively. Logistic regression was used to evaluate potential cognitive and behavioral predictors of later SIB.

**Results:** SIB was reported for 32% of participants at age 24 months. The first logistic regression model included sex, Mullen Early Learning Composite, Vineland Adaptive Behavior Composite, and endorsed SIB and stereotypy from the RBS-R. The overall model significantly predicted later SIB (χ2 = 31.2, p < .001, R2pseudo = 0.22). Of individual predictors, only Mullen ELC score and SIB at 12 months were significantly associated with SIB at age 24 months. Next, a second model excluding Vineland ABC and stereotypy endorsed scores was fit. The overall model was significant (χ2 = 34.8, p < .001, R2pseudo = 0.19). For participants who exhibited SIB at 12 months, the odds of engaging in SIB at 24 months increased by 93%. Odds of SIB decreased by 3% for each unit increase in Mullen ELC score. Follow-up analyses indicated that of Mullen subscales, Receptive Language and Visual Reception in particular were most strongly associated with later SIB.

**Discussion:** The purpose of this study was to evaluate behavioral characteristics at age 12 months predicting SIB at 24 months among infants at high familial risk for ASD. The logistic regression results indicated that presence of SIB and lower intellectual functioning at age 12 months significantly predicted later SIB. In contrast with previous studies of potential risk factors and correlates for SIB, stereotypy did not significantly predict SIB in our sample. This may indicate a developmental relationship between stereotypy and SIB which, early in life, qualitatively differs from that observed in older individuals. Identifying risk factors for the early development of SIB, and understanding the trajectory of this behavior disorder in early childhood, has the potential to inform prevention and early intervention programming.

**References/Citations:**

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**Paper Title:** A within Cohort Analysis of Predictors for Self-Injurious Behaviour and Self-Restraint in Autism Spectrum Disorder; Towards a Hypothesis of Impaired Behavioral Control

**Authors:** Caroline Richards¹, Louise Daniel⁹, Chris Oliver³

**Introduction:** There are few studies investigating behavioural correlates of self-injury, and the associations between self-injury and self-restraint in children and adults with Autism Spectrum Disorders. In this study, the prevalence of self-injury and self-restraint were established in a large cohort with ASD and behavioural correlates of both self-injury and self-restraint were identified. Predictive models for both behaviours were developed to explain the occurrence of self-restraint.

**Methods:** Teachers and key workers of 208 children and 216 adults with ASD (mean sample age = 24.10, range 6-61) completed questionnaires relating to the presence of self-injury and self-restraint behaviours. Information was gathered on demographic

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characteristics, health conditions, overactive and impulsive behaviour and repetitive and restricted behaviour. Analyses were conducted to identify predictors of both self-injury and self-restraint.

**Results:** Self-injury was found to be highly prevalent in both children (45.7%) and adults (49.1%) with ASD. Self-injury was associated with, and predicted by, the presence of lower ability, painful health conditions, repetitive and restricted behaviours and overactive and impulsive behaviour. Self-restraint was also found to be common in children (40.4%) and adults (42.6%), and was associated with, and predicted by the presence of self-injurious behaviour, repetitive and restricted behaviour and overactive and impulsive behaviour.

**Conclusions:** The implications of these findings are discussed in relation to a potential role for impaired behavioural inhibition and painful health conditions in the development and maintenance of self-injury and self-restraint in ASD.

**References/Citations:**

**Paper Title:** Self-Injurious Behavior and the Autisms: One Size Does Not Seem to Fit All

**Authors:** Allison Whitten$^{11}$, Mika Garrett$^{12}$, James W. Bodfish$^{11}$

**Introduction:** Clinically, "self-injurious behavior" (SIB) seems to manifest in diverse ways across cases; however, in research studies, this apparent phenomenologic variety is often overlooked. Phenotypic variability is a challenge for studies of mechanism and intervention, and therefore research that attempts to parse putative clinical heterogeneity is called for. The presence of intellectual disability (ID) and also other neurodevelopmental disorders such as autism spectrum disorder (ASD) increases the risk that SIB will develop. The objective of this study was to examine differences in the occurrence of SIB and differences in the relation of SIB to clinical correlates in samples of children with ASD subgrouped based on the presence or absence of ID.

**Methods:** 81 school-aged children (mean age 12.4 [SD 3.18]; 85% males) with a clinical diagnosis of ASD, who exceeded ASD current symptom cutoff scores (SCQ) and met research diagnostic criteria for ASD (ADOS, ADIR, & expert clinical opinion), were subgrouped based on nonverbal IQ (> 90; < 70) resulting in n = 30 cases with ASD + cognitive impairment (ASD+ID), and n = 41 cases with ASD and no cognitive impairment (ASD-only); 10 additional cases were excluded (below ADOS/ADI cutoff, or IQ between 70-90). All included cases were evaluated using a multi-measure / multi-method (parent rating scale, structured clinical interview, structured observation) deep phenotyping approach using a battery of repetitive behavior, social-communication, language ability, and psychopathology measures. In a subset of cases data from passive eye-tracking and EEG tasks were also administered as an initial effort to examine feasibility of tasks that could provide reliable biomarkers of phenotypic subgroups that emerged.

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Results: SIB occurred in the ASD+ID (63%) and the ASD-only (42%) subgroups, but severity was greater in the ASD+ID subgroup (p = .002). SIB was significantly correlated with the severity of stereotyped behavior \( (r = .53) \) and with expressive language ability \( (r = .71) \) only in the ASD+ID subgroup. SIB was significantly correlated with anxiety severity \( (r = .59) \), and OCD severity \( (r = .37) \) only in the ASD-only subgroup. The ASD subgroups could also be distinguished using selected eye-tracking and EEG metrics.

Pitfalls: Finding clinical measures that can be used across a wide range of cognitive and language abilities is challenging, and the measures used very likely have different degrees of psychometric strength in this regard.

Discussion: SIB occurs frequently in the context of autism but appears to have a different pattern of clinical correlates in cases with co-occurring ID (where it may be most associated with other repetitive behaviors and with language impairments) relative to those without general cognitive impairments (where it may be most associated with co-occurring psychopathology like anxiety and OCD). Differential patterns of clinical correlates may help to guide treatment development research. If so, this may support a shift towards a "personalized medicine" approach that could map on to the known clinical heterogeneity of both SIB and autism.

Supported by NIMH R01 073402; NICHD P30 HD15052

Paper 7 of 8

Title: Identifying environmental influences on SIB shown by boys with fragile X syndrome

Authors: Scott S. Hall, Rebecca P. Barnett, Kristin M. Hustyi

Affiliation: Stanford University

Introduction: A significant proportion of individuals with fragile X syndrome (FXS), the most common known form of inherited intellectual disability, display self-injurious behavior (SIB) that can pose a significant threat to the child’s health and safety. Toward the goal of advancing fragile-X specific behavioral treatments for SIB in FXS, we examined the influence of social-environmental factors on SIB shown by adolescent boys with FXS.

Method: Twenty boys with FXS, aged 10 to 18 years, received an experimental functional analysis consisting of seven conditions: Ignore, Attention, Tangible, Academic Demand, Social Demand, Transitions and Play. The Social Demand condition (social conversation requiring eye contact) and the Transition condition (moving from one moderately preferred activity to another) were included to address the distinctive behavioral profile of FXS.

Results: Results showed that in 17 of 20 cases, SIB was influenced by social-environmental factors. Nine of the 20 children also showed aggressive behaviors in the functional analysis. There were no differences between those who showed SIB only and those who showed SIB and aggression.

Discussion: These data indicate that pharmacological treatments to reduce problem behaviors in FXS are unlikely to be successful unless social-environmental influences maintaining these behaviors are considered.
**Title:** Delineating Subtype of Self-Injurious Behavior Maintained by Automatic Reinforcement: Treatment Outcome Data

**Authors:** Jennifer Zarcone, Griffin Rooker, Louis Hagopian

**Affiliations:** Kennedy Krieger Institute, Johns Hopkins School of Medicine, Baltimore, MD Zarcone@kennedykrieger.org

**Introduction:** Self-injurious behavior (SIB) is maintained by automatic reinforcement for roughly 25% of individuals with SIB. Automatically reinforced SIB typically has been considered a single functional category, but we have proposed a model for subtyping automatically reinforced SIB based on functional analysis outcome. We applied this model to archival treatment data obtained on our inpatient unit to determine if treatment outcome, with regard to type of treatment and resistance to treatment, would correspond to SIB subtype.

**Methods:** The current study involved an analysis of the treatment data from 39 individuals with automatically reinforced SIB and a comparison group of 13 individuals with socially reinforced SIB. Automatically reinforced SIB was categorized into 3 subtypes based on patterns of responding in the functional analysis and the presence of self-restraint. Treatment was identified as effective if there was an 80% or greater reduction in SIB.

**Results:** Results indicated that reinforcement alone (and/or with extinction) was effective for individuals with socially maintained SIB (85%) and Subtype 1 automatic SIB (67%). For individuals with Subtype 2, who engage in high rates of SIB across functional analysis conditions, treatment with reinforcement alone was ineffective. In fact, for all 15 participants with this subtype, additional interventions were required for the treatment to effectively reduce behavior (e.g., blocking, punishment, restraint, or all of the above). Even with these much more intensive interventions, only 54% of the participants had a reduction in behavior of 80% or greater.

**Discussion:** The level of differentiation in the functional analysis serves as an excellent model for predicting treatment type and outcome. Individuals with Subtype 1 had patterns of behavior and treatment outcomes much more similar to individuals with socially-maintained SIB. We have expanded our model to published studies using the same subtype model. Results showed the exact same patterns as our clinical data, supporting the idea that this model is robust and can be used as a means of identifying responders (and possibly nonresponders) to treatment.

**Key References:**

Supported, in part, by Eunice Kennedy Shriver NICHD Grant No: 76653
**Symposium Title:** Attention in Autism and other Neurodevelopmental Disorders from Infancy to Adulthood

**Chair:** Bridgette Tonnsen

**Discussant:** Elisabeth Dykens

**Overview:** Atypical attention is well documented in neurodevelopmental disorders and is posited to contribute to multiple areas of impairment, including social communication, anxiety, and behavior problems. Recent advances in experimental eye movement technologies have improved researchers' capacity to examine these fine-grained associations, potentially improving characterization of phenotypes and treatment targets. This international symposium features early career researchers who integrate experimental technology to investigate attention-related mechanisms of neurodevelopmental impairment across multiple syndromes and domains. Tonnsen and colleagues examine integrated behavioral and heart-defined attention as potential markers of prodromal autism risk in infants with fragile X syndrome (FXS) and infant siblings of children with autism spectrum disorder (ASD). Lee and colleagues characterize complex associations between attention and both linguistic and social development in adolescents and adults with ASD. Crawford and colleagues define cross-syndrome differences in the intersection of anxiety and attention across adolescents with ASD, FXS, Cornelia de Lange syndrome, and Rubinstein-Taybi syndrome. Collectively, these studies unveil novel information about the intersection of attention and neurodevelopmental phenotypes, while also highlighting the challenges, benefits, and diverse applications of eye-movement research in special populations.

**Paper 1 of 3**

**Paper Title:** Cross-Syndrome Attention in Infants at Risk for Autism

**Authors:** Bridgette Tonnsen, John Richards, Jane Roberts

**Introduction:** Characterizing early predictors of autism facilitates earlier identification, diagnosis, and treatment. Aberrant attention orienting is one of the earliest identified predictors of autism and may play an integral role in developmental cascades that contribute to impairments. However, the emergence of atypical attention in infants at risk for autism is poorly understood. Previous studies of attention in high risk infants have largely focused on infant siblings of children at risk for autism (ASIBs), who exhibit 10-20 times higher rates of autism diagnoses than the general population (Ozonoff et al., 2011). Far fewer studies have been conducted in syndromic samples at elevated risk for autism, including infants with fragile X syndrome (FXS), the leading heritable cause of autism. Comparing early risk factors in ASIBs and infants with FXS may inform the generalizability of early autism markers, as well as potential syndrome-specific pathways.

The present study used a cross-syndrome, multimethod approach to examine the generalizability and mechanisms of attention orienting in infants at high risk for autism. We used a behavioral orienting task with concurrently-measured heart-defined sustained attention to address the following questions: (1) Do behavioral and heart-defined markers of attention orienting differ across FXS, ASIBs, and low risk (LR) controls? (2) Within FXS, does attention predict clinical autism risk at 12 months of age?

**Methods:** Participants included infants with FXS assessed at 9 (n=13) and 12 (n=14) months of age, matched to same-age ASIBs (9m n=19; 12m n=18) and LR controls (9m n=18; 12m n=21). Approximately 65% of participants were assessed at both time points. FXS data were also contrasted to MA-matched LR controls (9m n=12; 12m n=18). Infants were administered the gap-overlap task, a commonly used paradigm to measure flexibility of attention orienting. Primary behavioral variables included

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latency to disengage attention from competing stimuli ("attention disengagement") and overall looking time towards task stimuli. We also measured concurrent heart-defined sustained attention, a well-validated physiological orienting response characterized by decelerated heart rate (e.g. Richards & Casey, 1991). Primary physiological variables included proportion of time in sustained attention and variability of heart rate during sustained attention. Clinical autism risk at 12 months was measured using the Autism Observation Scale for Infants.

**Results:** Data were analyzed cross-sectionally at 9 and 12 months using nonparametric Wilcoxon-Mann-Whitney tests and Spearman correlations. Results indicated persistent, early-emerging behavioral orienting deficits in FXS that increasingly deviated from MA controls between 9 and 12 months. The magnitude of abnormality was greater in FXS than ASIBs. Biobehavioral associations also differed across groups. Whereas behavioral attention positively correlated with heart-defined sustained attention in LR controls, the FXS group exhibited a paradoxical pattern of greater behavioral attention in participants with poorer sustained attention, and ASIBs did not exhibit any significant biobehavioral associations. Clinical autism risk was predicted by behavioral attention disengagement, but not physiological variables, across both FXS and ASIB groups.

**Discussion:** In one of the first cross-group studies of prodromal autism risk in high-risk infants, we observed shared behavioral attention patterns in FXS and ASIBs that appear to be subserved by distinct physiological processes. These data suggest that abnormal attention may relate to different physiological mechanisms across high-risk FXS and ASIB groups, potentially necessitating different screening and treatment protocols. This work warrants further study of cross-syndrome attention mechanisms and developmental processes in infants at risk for autism.

**References/Citations:**


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**Paper 2 of 3**

**Paper Title:** Looking and Language in Autism Spectrum Disorder: A Comprehensive Investigation

**Authors:** Michelle Lee⁴, Nell Heckel⁵, Kritka Nayar⁴, Abigail Hogan-Brown⁴, Peter Gordon⁵, Gary Martin⁷, Molly Losh⁵

**Introduction:** Converging evidence suggests that individuals with autism spectrum disorder (ASD) display atypical patterns of visual attention (1-3) that predict greater social impairment (2). Therefore, characterizing visual attention in ASD may provide key insights into the cognitive mechanisms underlying social-linguistic processes in this population. However, research has not examined visual and attentional processes during and across communicative acts, nor how these influence language output. This study comprehensively investigated relationships between visual attention and language in individuals with ASD during two narrative tasks varying in structure and a Rapid Automatized Naming task.

**Methods:** Thirty-four individuals with high functioning ASD (IQ>80) and 24 age-matched controls completed three language tasks presented on an eye-tracker: a structured wordless picture book, Frog, Where Are You?(4), a semi-structured task employing images from the Thematic Apperception Test (TAT)(5), and a Rapid Automatized Naming (RAN) task. RAN involves quickly naming

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letters, numbers, colors, and objects and has been used extensively to predict reading fluency and phonological processes (6). To assess relationships between visual attention and language during this task, we calculated the eye-voice span (EVS), or the number of items ahead the eyes are while articulating an item. During the picture book task, participants narrated each page as it was presented on the eye-tracker. For the TAT task, individuals viewed six images from the TAT for eight seconds each. After each image was removed, participants were asked to tell a story about the image. Visual attention for narrative tasks was quantified by proportion of fixations to different aspects of the images; narrative quality was measured using computational linguistic measures of semantic content.

Results: During the RAN task, participants with ASD demonstrated smaller EVS than the control group (p < .01), suggesting that individuals with ASD required greater attentional resources to complete the task. The ASD group’s attention to social stimuli was correlated across narrative tasks (r > .4). However, in contrast to prior passive viewing studies, individuals with ASD did not differ from controls in their attention to social aspects of the stimuli. Rather, they demonstrated decreased attention to background elements during their narration (p < .05); greater attention to non-social stimuli was positively correlated with semantic quality across narrative tasks in both groups (r > .4).

Discussion: This study is the first to assess the influence of visual attention on language production across multiple contexts. Findings highlight the importance of measuring language and visual attention simultaneously. Individuals with ASD demonstrated reduced automaticity on the RAN task, which assesses critical precursors to more complex language usage. This lack of automaticity may lead to downstream language deficits. In contrast to passive viewing tasks, individuals with ASD directed their attention to social aspects of stimuli when explicitly directed to narrate; however, they differed in attention to key background elements, resulting in narratives less rich in semantic content. Overall, results lend further evidence to the link between atypical visual attention and language and social deficits in ASD, highlighting potential avenues of intervention and further investigation of ASD’s etiology.

References/Citations:
- Mayer (1969). Frog, where are you?
Paper Title: Visual Preference for Social versus Non-Social Stimuli in Children and Adults with Neurodevelopmental Disorders

Authors: Hayley Crawford, Joanna Moss, Chris Oliver, Natasha Elliott, Giles Anderson, Joseph Mc Cleery

Introduction: Recent research has identified differences in relative attention to competing social versus non-social video stimuli in autism spectrum disorders (ASD). However, it is yet to be investigated whether the extent to which the stimuli move towards participants, influences attentional allocation. In addition, existing research has highlighted distinct patterns of visual attention to social stimuli in individuals with other neurodevelopmental disorders, such as Williams syndrome, which appear consistent with the profiles of social behaviour displayed in this group. Whether such patterns exist for individuals with other neurodevelopmental disorders associated with subtle differences in social behaviour has yet to be investigated.

Methods: In Study 1, adolescents with ASD (n = 16) and control participants (n = 16) were presented with social and non-social video stimuli in directed and non-directed formats whilst their eye movements were recorded. In Study 2, this same paradigm was employed with individuals with Fragile X (n = 15), Cornelia de Lange (n = 14), and Rubinstein-Taybi syndromes (n = 19). Total looking time for directed versus non-directed social and non-social video stimuli (attention maintenance), and time taken to fixate on each stimulus type (attention priority), were analysed in each study.

Results: Consistent with previous studies, children with ASD demonstrated reduced attention maintenance for social versus non-social videos during the directed condition but this did not extend to the non-directed condition. Individuals in the three genetic syndrome groups showed similar attention maintenance, but differences in attentional priority, for directed social stimuli. Specifically, participants with Cornelia de Lange syndrome took longer to fixate to directed social stimuli than participants with Fragile X or Rubinstein-Taybi syndromes. In the Fragile X group, a positive relationship emerged between dwell time to directed social stimuli and social anxiety.

Discussion: The main findings across two studies indicate that those with and without ASD demonstrated differences on a relatively coarse measure of overall dwell time to social versus non-social videos (Study 1), whilst the nuanced measure of time taken to initially orient to social and non-social stimuli highlighted differences between groups exhibiting more subtle differences in their social presentation (Study 2). Critically, the differences observed between the groups are each consistent with previously documented differences in their respective social phenotypes. Taken together, these results provide further support for the high potential of relatively simple eye-tracking measures of visual attention to social versus non-social stimuli to highlight differences across populations with associated atypical socio-behavioural profiles.

References/Citations:


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8 Centre for Research in Psychology, Behaviour & Achievement, Coventry University, UK
9 Cerebra Centre for Neurodevelopmental Disorders, School of Psychology, University of Birmingham, UK
10 Institute of Cognitive Neuroscience, University College London, UK
11 School of Psychology, University of Birmingham, UK
12 School of Psychology, Oxford Brookes University, UK
13 Center for Autism Research, Children's Hospital of Philadelphia
Symposium Title: Parenting and Longitudinal Development in Children with or without Intellectual Disability

Chair: Jan Blacher

Discussant: Cameron L. Neece

Overview: Certain childhood developmental competencies have been extensively linked to later academic, social, mental health, and functional outcomes in adolescence and adulthood. For example, children who exhibit high levels of behavior problems are more likely to underachieve academically, commit a crime later in life, and develop substance use problems. Children with language delays or communication difficulties are likely to have difficulties with literacy, employment, and attention problems. Lastly, research indicates that emotion dysregulation and sensitivity predict peer relationship quality and psychopathology prevalence.

Research has demonstrated that children with intellectual disability (ID) are more likely to show delayed communication skills, poor emotion regulation, and high rates of behavior problems in school and at home. In addition, researchers have presented evidence that individual genetic variation confer susceptibility to environmental contexts associated with these fundamental competencies, such as emotional lability, impulsivity, and attention problems. As these core skills underlie many other functional outcomes, risk factors related to these fundamental skills have extensive repercussions through priming specific children to experience difficulties throughout childhood and into adulthood. This symposium will explore the multiple contributions of parenting to the longitudinal development of these foundational skills for children across the full range of cognitive ability.

Parenting has been linked to the development of the foundational skills of language, emotion regulation, and positive behavior, primarily through cross-sectional designs. This symposium will examine changes in child competencies over time, as predicted by specific characteristics of parents and parenting behavior. These findings are particularly powerful for children who are at high risk for negative outcomes due to biological risk and/or developmental delays, for which research suggests environment is an especially influential variable. All studies in this symposium have strong implications for childhood interventions through fostering protective parenting factors to promote positive outcomes for all children.

All papers in this symposium draw on a longitudinal study of development in children with or without intellectual disability. The first paper in this symposium will illuminate the relationship between parenting stress, parent perceptions, and childhood behavior problems for children with or without ID over time using a moderated mediation model. The second paper examines how the specific language input parents model for children predicts the development of complex language skills, such as use of multi-clause sentences, in children with or without ID. Finally, the third paper analyzes the impact of genetic susceptibility and of cognitive ability on emotion regulation development in the context of positive and negative parenting.

Paper 1 of 3

Paper Title: Parenting Stress and Child Behavior Problems in Children with or without Intellectual Disability: The Mediating Role of Maternal Expressed Criticism

Authors: Barbara Caplan, Willa Marquis, Bruce L. Baker

1 University of California, Riverside
2 Loma Linda University
3 University of California, Los Angeles
Introduction: The relationship between parenting stress and child problem behavior has been well documented in investigations of typically developing (TD) children (Crnic, Gaze, Hofmann, 2005), as well as those with developmental disabilities (Davis & Carter, 2008; Baker et al., 2003). This relationship is of particular concern for families of children with intellectual disability (ID), who demonstrate elevated levels parenting-related stress, as well as increased levels of child behavior problems (Baker, Blacher, Crnic, Edelman, 2002) relative to TD children. Consistent with transactional models of child development (Sameroff & Chandler, 1975), parenting stress and child behavior problems have been shown to have a mutual influence on one another (Neece, Green & Baker, 2012; Baker et al., 2003), resulting in escalating or de-escalating relationships over time. Some evidence suggests that parenting stress influences child behavior through parenting interactions with their children (Crnic & Low, 2002). However, multiple pathways from parenting stress to child behavior problems may be at play. For example, parental expressed emotion toward their children is proposed to reflect "toxic family stress", and is also associated with child behavior problems over time (Peris & Miklowitz, 2015). Further, though the relationship between parenting stress and behavior problems has been established for children with and without ID, no known investigations have examined whether mediating processes may differ by child disability status.

The present study investigated status group (ID vs. TD) differences in maternal stress, maternal expressed emotion, and child behavior problems. Further, this study tested a novel proposed mediator of the relationship between parenting stress and child behavior problems (maternal expressed emotion), and examined whether child disability status (ID vs. TD) moderates this mediating relationship.

Methods: Participants were 133 families from a longitudinal study of children with ID (n=46) or TD (n=87). Children and their families were assessed at child ages 7, 8 and 9 years either at a university laboratory or in the home. At the child age 7 visit, parents were interviewed regarding family demographic information and completed questionnaires regarding their parenting-related stress using the Family Impact Questionnaire (Donenberg & Baker, 1993). At child age 8, parents provided a brief (two-minute) speech sample, coded using an EE rating system adapted for parents of children with ID. Parent then rated their child's behavior problems at child age 9 using the well-validated Child Behavior Checklist (Achenbach & Rescorla, 2001).

Results: Analyses revealed that mothers of children with ID reported greater levels of parenting stress than mothers of TD children (t= 5.97, p<.001), but did not exhibit higher levels of expressed criticism when talking about their child. As expected, children with ID exhibited greater levels of externalizing behavior than TD children (t=3.87, p<.001). Mediation hypotheses were analyzed using PROCESS for SPSS (Hayes, 2012). Results indicated that the overall mediation effect of maternal expressed criticism was not significant. However, a significant moderated mediation effect was found such that maternal criticism was a significant mediator of parenting stress and child behavior problems only for TD children.

Discussion: Implications for developmental theory and parenting interventions will be discussed.

References/Citations:

Paper Title: Characteristics of Parent Language Input: Predicting Child Complex Syntax in Children with and without Developmental Delays

Authors: Christine T. Moody, Bruce L. Baker

Introduction: Research documents the facilitative effects of language input and positive parenting strategies on early communication skills, including first words and vocabulary, in children with and without intellectual disability (ID; Warren & Brady, 2007; Hart & Risley, 2002). However, few studies have examined the continued contribution of parent language input to increasingly complex language skills through the preschool years. Research indicates that individual differences in child language complexity are related to the proportion of complex speech modeled by parents (Huttenlocher et al., 2002). Further, overall quantity and diversity of parent complex speech partially mediates the relationship between SES and child language outcomes (Huttenlocher et al., 2010). Despite these initial studies, current research is limited to typically developing children. It is necessary to examine how language input serves to support the acquisition of complex language in children with developmental delays, who are at risk for poor outcomes.

Methods: Subjects include families (n=208) who participated in a longitudinal study of development in children with and without ID. This study will use data coded from videotaped and transcribed mother-child interactions at ages 3, 4, and 5. Child complex syntop will be coded using the Index of Productive Syntax (IPSyn; Scarborough, 1990). Three characteristics of parent language input will be measured: quantity, utterance function (e.g. comments, commands), and complexity of speech. To predict the rate of growth in complex syntax over the preschool period, multiple regression analyses will be conducted to determine the predictive power of parent language input characteristics, while controlling for earlier child syntax at age 3. Further, we will examine if these characteristics differentially impact complex language learning for children with developmental delays as compared to typically developing children.

Results: Transcription and coding are ongoing. With a preliminary sample (n=30), children with developmental delays use less complex language at age three (M=39.27, SD=15.65) than do typically developing children at age three (M=62.20, SD=9.35), t=4.87, p<.001. This discrepancy across groups is maintained at age four as well, t=3.19, p=.004, (DD: M=59.67, SD=15.76; TD: M=74.27, SD=8.11. Children with developmental delays also show more variability in IPSyn scores at age 4, ranging from 30 to 82 points, than did typically developing children, whose scores ranged from 60 to 87. Further, with the current sample size, child language complexity at age 4 is not correlated with cognitive ability in children with developmental delays, r=.327, ns, suggesting that there are other influential variables present.

Discussion: Literature suggests that parents play a critical role in early child language learning. However, much of the current research focuses on children under age 3, uses limited outcome variables (e.g. vocabulary), and only includes typically developing children. It is difficult to generalize these results to children with developmental delays who may have unique learning needs. Information about aspects of parenting behaviors that are influential to child development would create knowledge that could be translated to recommendations for parents as their child grows.
References/Citations:


Paper 3 of 3

**Paper Title:** Growth Curve Models of Child Emotion Dysregulation as Predicted By Cognitive Ability and Serotonin Transporter Genotype-Parenting Interactions

**Authors:** Amanda Norona, Irene Tung, Steve Lee, Jan Blacher, Bruce L. Baker

**Introduction:** Given that individual differences in emotion regulation (ER) predict numerous long-term psychological, emotional, and physical outcomes, there has been considerable effort to identify the developmental processes underlying ER. Two broad factors are central to the development and maintenance of ER: influences external to the individual, such as family modeling and parenting practices, versus internal influences, such as temperament and neurobiological reactivity (Calkins, 1994). More recently, genetic association studies have examined the biological substrates of ER. For example, the promoter polymorphism of the serotonin transporter gene has been associated with ER, although the evidence is inconsistent: whereas some evidence suggests that the low-expression short allele was associated with emotion dysregulation, particularly in the presence of stressful environments (e.g., Hariri & Holmes, 2006), other evidence is suggestive of potential differential susceptibility, where poor ER was predicted from environmental adversity, but the same genotype conferred optimal ER in the presence of environmental enrichment (e.g., Hankin, et al., 2011).

Reflecting this important dissociation, we examined how internal and external factors (i.e., genotype, cognitive ability, and parenting) affected the trajectory of emotion dysregulation. We hypothesized that parenting behavior would significantly predict dysregulation such that negative parenting would predict increases in dysregulation and positive parenting would produce decreases. In addition, we predicted that serotonin transporter genotype would significantly moderate prospective predictions of dysregulation from parenting behavior consistent with differential susceptibility model, such that the parenting effects would be exaggerated for those with the low-expression genotype. Lastly we predicted that cognitive ability would predict initial levels of emotion dysregulation but not its trajectory.

**Methods:** Participants were 96 families in the Collaborative Family Study, a longitudinal study of children with and without developmental delays. Child emotion dysregulation and positive and negative parenting were coded from mother-child lab interactions. Child dysregulation, defined as intense or frequent expressions of emotion inappropriate to lab tasks, was measured at child ages 3, 4, 5, and 6. Early positive and negative parenting was measured at child ages 3 and 4. Child cognitive ability was measured at age 5. We used latent growth curve modeling to model yearly change in dysregulation from age 3 to 6. We examined the main effect of cognitive ability and whether serotonin transporter genotype moderated relationships between behavior codes of positive and negative parenting and the developmental trajectory of child dysregulation. Our genotype variable was a composite of 5-HTTLPR and rs25531; individuals with two low-expression alleles (S/S, S/LG, LG/LG) were coded as 1, all others as 0.
**Results:** Latent growth curve modeling revealed that a negative quadratic slope best captured change in child emotion dysregulation. Negative parenting predicted an increase in the quadratic slope of child dysregulation over time, and more so for those with the low-expression genotype. In addition, positive parenting predicted a decrease in the quadratic slope of child dysregulation over time, only for those with the low-expression genotype. GxE did not predict baseline levels of emotion dysregulation at age 3, only the change over time. Child cognitive ability predicted the rate of change of emotion dysregulation, but not baseline levels.

**Discussion:** Overall, these preliminary findings were suggestive of the differential susceptibility hypothesis, as positive and negative parenting differentially affected the trajectory of child emotion dysregulation.

**References/Citations:**
- Overall, these preliminary findings were suggestive of the differential susceptibility hypothesis, as positive and negative parenting differentially affected the trajectory of child emotion dysregulation.
- References
- Hankin et al. (2011). Differential susceptibility in youth: evidence that 5-HTTLPR x positive parenting is associated with positive affect 'for better and worse' Transl. Psychiatry, 1(10), e44.