

Symposium Title: What is the need, and what do we do? Innovations in assessing and treating families of individuals with neurodevelopmental disorders during COVID-19 and beyond.

Chair: Bridgette Kelleher¹ and Anne Wheeler² (co-chairs)

Discussant: Elizabeth Jalazo³

Overview: The COVID-19 pandemic has caused global shifts to family and community life, particularly for families affected by neurodevelopmental disorders whose stress and service needs were elevated even before the pandemic. At the same time, the pandemic has caused widespread shifts in how researchers and clinicians can support families, increasing reliance on methods and technologies that can monitor and treat high-risk families remotely. The four manuscripts in this symposium each capture a unique approach to assessing or treating the needs of families affected by neurodevelopmental disorders during COVID-19. Presentations feature speakers from a variety of career phases, institutions, and disciplines. The first presentation (Wheeler) uses a large national survey to characterize how COVID-19 has impacted families of children with NDDs, relative to those without NDDs, including by using a novel quantitative approach – best-worst scaling – to quantify relative worries across groups. The second presentation (Kelleher) zooms in to follow the daily experiences of families of children with rare neurogenetic syndromes who were recruited from an ongoing longitudinal study. She examines how changes in mental health symptoms and caregiving experiences relate to caregivers' momentary experiences of stress, assessed using ecological momentary assessment "pings" sent to caregivers' smart phones. The third presentation (Panjwani) shifts focus to how COVID-19 has impacted a specific area of concern for many NDD families: food, nutrition, and food insecurity. She specifically examines the experiences of children with autism and how food insecurity relates to family experiences. The final presentation (Bucher/Shannon) asks the critical question – so what do we do to help? In this talk, graduate students Bucher and Shannon describe an ongoing telehealth intervention study in which caregivers of children with Angelman syndrome were randomized to receive personalized, telehealth-based mental health treatment, behavioral coaching, or both treatments combined. They describe how families responded to treatment during COVID-19 and how this model may be broadened to other NDD populations and therapies. Together, these presentations will provide a snapshot of the unique challenges faced by families of children with NDDs during the COVID-19 pandemic and inform how we as researchers and clinicians can best support them during this challenging time. The symposium will conclude with a discussion led by Dr. Elizabeth Jalazo, a clinician and parent of a child with a neurodevelopmental disorder.

Paper 1 of 4

Paper Title: Understanding the impact of the COVID-19 pandemic on families of children with disabilities: Results from a nationwide survey

Authors: Anne Wheeler², Holly Peay², Katherine Okoniewski², Katherine Porter², Kay Hwang²

Introduction: Families of children with disabilities have been especially impacted by substantial shifts in working environments, implementation of virtual learning, closing of childcare centers, and a lack of intervention service provision during the COVID-19 pandemic (1). Meeting the needs of children with behavioral, communication, cognitive, or health impairments within a systemic global crisis becomes additionally complicated with the potential of increases in challenging behaviors and needs (2). Caregivers quickly became interventionists taking on the role of educator and support service provider amongst closings, virtual implementation of services, and transitions home from residential care facilities (3). The goals of this study were to capture the needs and concerns of families of children with and without disabilities during the initial months of the pandemic. Specifically,

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we focused on caregiver mental health and factors that families found to be the most worrisome related to COVID-19, and differences based on whether they had children with disabilities.

Methods: Parents were recruited directly from active research studies by the team, through advocacy organization listservs, and through social media outlets. Respondents completed an electronic survey between April and June, 2020. The survey assessed both caregiver factors – such as mental health and stress utilizing the Depression, Anxiety and Stress Scale (4)– as well as child factors – such as challenging behaviors and coping skills. Comparisons of parents with and without children with special needs or health conditions impacting daily life or learning (as self-reported by participants) were assessed via t-tests and chi-square analysis. Participants completed a Best-Worst Scaling (BWS) case 1 activity to asking them to respond to 15 choice sets prioritize worries for themselves and their families since the COVID-19 pandemic began. Worry variables were developed based on the literature and anecdotal reports.

Results: Preliminary results indicate parents of children with disabilities reported significantly more stress ($p = 0.003$) and depression symptoms ($p < .001$) than parents of children without special needs. BWS results show that all parents reported their greatest worries were around COVID-19 related risks to their child or other family members. Families with a child with a disability prioritized school or daycare closures and the loss of services or activities for their child due to COVID-19 much more than parents whose children without disabilities. Between 44% and 56% of parents of children with special needs report increases in aggression, anxiety, hyperactivity, distractibility, crying, tantrums, and clinginess since COVID-19 pandemic began. In contrast only 10%–20% of parents of children the same age but without disabilities report increases in these behaviors.

Discussion: The COVID-19 pandemic has had a significant impact on families all over the world. This survey also suggested the pandemic has had a disproportionately negative impact among children with disabilities. Parents of children with disabilities, whose baseline stress levels were already higher than those whose children do not have disabilities, may be experiencing exacerbated mental health risks because of the pandemic. Children with disabilities were significantly more likely to exhibit an increase in behavior challenges, and families face added stressors such as their child's increased risk for needing non-COVID-19 related medical care and a loss of relied upon services. It is important as researchers and clinicians dedicated to supporting individuals with disabilities and their family, that we recognize the unique vulnerabilities experienced by these populations.

References/Citations:

1. Liu JJ, Bao Y, Huang X, Shi J, Lu L. Mental health considerations for children quarantined because of COVID-19. *The Lancet Child & adolescent health*. 2020;4(5):347-9.
2. Courtenay K, Perera B. Covid-19 and people with intellectual disability: Impacts of a pandemic. *Irish Journal of Psychological Medicine*. 2020.
3. Rose J, Willner P, Cooper V, Langdon PE, Murphy GH, Kroese BS. The effect on and experience of families with a member who has Intellectual and Developmental Disabilities of the COVID-19 pandemic in the UK: developing an investigation. *International Journal of Developmental Disabilities*. 2020;Early Access.

Paper 2 of 4

Paper Title: Increased caregiving burden due to COVID-19 uniquely predicts mental health and stress in a prospective cohort of mothers of children with rare syndromes

Authors: Bridgette Kelleher¹, Wei Siong Neo¹, Nicole Witthuhn^{1, 4}, Riley Felicicchia¹, Tyra Protho¹, Cameron Dartis¹, Dan Foti¹, Sean Lane¹

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Introduction: The COVID-19 pandemic has substantially impacted families worldwide. For caregivers of children with special needs – such as neurogenetic syndromes (NGS) associated with intellectual disability – the impacts are likely magnified, particularly given the higher caregiving burden, medical complexities, and SARS-CoV-2 related risk factors associated with these conditions. Much of the global scientific focus on COVID-19 to date has understandably focused on tracking and minimizing the physical health consequences of the virus. However, much less is known regarding the psychosocial impacts of COVID-19, particularly on caregivers. The present study addresses this gap by examining how the COVID-19 pandemic has impacted daily experiences of 50 NGS caregivers who are participating in a 7-week ecological momentary assessments study during the pandemic. Importantly, these caregivers were recruited from an ongoing longitudinal study that started prior to COVID-19, enabling us to compare symptoms pre- and post- COVID-19 onset. Our goals were twofold. First, we aimed to evaluate how self-reported COVID-19 stressors -- such as increased caregiving responsibilities, stress due to COVID-19, and social isolation -- moderated changes in caregiver mental health changes from pre-pandemic baseline. Second, we aimed to characterize how these COVID-19 stressors directly impact daily life by examining their association with daily fluctuations in caregiver stress and mood, assessed throughout the day by “pings” sent to caregiver smartphones.

Methods: Participants were caregivers of children with NGS enrolled in the Purdue Early Phenotype Study. Families were primarily white (96%), and 86% reported family income above \$50,000. Child probands were diagnosed with Angelman (n=13), Down (n=9), fragile X (n=4), Prader Willi (n=9), and Williams (n=15) syndromes. Child ages ranged from 1-14 years ($M = 4.5$) at the time of their first post-COVID-19 assessment. Here, we focus on data collected through (1) an introductory survey on general functioning during COVID-19, which included the Depression, Anxiety, and Stress Scale (DASS-21; Lovibond and Lovibond, 1995) and questions related to COVID-19 related stress, isolation, and childcare responsibilities and (2) brief ecological momentary assessments (EMA), collected 4x per day via a smartphone app, that measured positive and negative affect using the I-PANAS-SF (Thompson, 2007) and measured stress using the question, “How STRESSED are you feeling right now?” (sliding scale; 0 = “Not at all” and 100 = “Extremely”). The EMA portion of the study is ongoing; here, we focus on the first 7 days of EMA data (1,060 observations with 24.1% missing data).

Results: Prior to COVID-19, the majority of caregivers reported DASS-21 symptoms in the “normal” range. After COVID-19 onset, nonparametric sign tests indicated significant increases in stress (56% increased; $d = .29$, $p = .044$) and a small but non-significant increase in depression symptoms (36%; $d = .16$, $p = .122$). Changes in anxiety were more variable across participants and, as a whole, did not increase (28%; $d = -.07$, $p = .856$). To identify potential moderators of increased symptoms, we conducted three linear regression models predicting each DASS-21 domain from COVID-19 stressors (change in caregiving, current level of caregiving, family social isolation), controlling pre-COVID-19 DASS-21 scores. Across models, change in caregiving responsibility, but not general level of caregiving responsibility, predicted depression ($p = .007$), anxiety ($p = .001$) and stress ($p = .006$). There also was a trend toward higher depression ($p = .075$) and stress ($p = .076$) among those with greater family isolation.

We next used nested mixed effects models to test how increased caregiving burden due to COVID-19 related to daily patterns of stress, positive affect, and negative affect collected via EMA. We used restricted maximum likelihood estimation for all models and Satterthwaite approximation for computing degrees of freedom. Intraclass correlation coefficients indicated that stress and affect varied considerably within and across caregivers, with affect demonstrating more within-person stability. Caregiver ratings of stress and affect fluctuated across the week, with significantly lower stress on weekends (Saturday and Sunday) and greatest negative affect mid-week (Thursday). Stress and positive affect were both higher in the mornings than evenings. Participants experiencing greater changes in caregiving responsibility reported higher mean levels of stress ($p < .001$) and negative affect ($p = .002$) and lower positive affect ($p = .016$) across observations.

Discussion: Our findings suggest that the majority of caregivers in our ongoing longitudinal study are experiencing increased stress relative to pre-pandemic baseline, and changes in caregiving burden due to COVID-19 appears to be a primary factor in predicting both changes in mental health functioning and daily fluctuations in stress and mood. Our ongoing work will continue to follow these caregivers through the pandemic to examine how health behaviors (e.g. exercise, substance use), social systems

(educational services, social relationships), child symptoms, and needs may moderate individual outcomes. However, our initial results suggest that higher caregiving burden due to COVID-19 is a particularly salient risk factor for NGS caregivers that should be targeted in public health policy and intervention.

References/Citations:

1. Lovibond, S. H. and Lovibond, P. F. (1995) *Manual for the depression anxiety stress scales*. Sydney: Psychology Foundation.
2. Thompson, E. R. (2007) 'Development and validation of an internationally reliable short-form of the Positive and Negative Affect Schedule (PANAS)', *Journal of Cross-Cultural Psychology*, 38(2), pp. 227–242.

Paper 3 of 4

Paper Title: COVID-19 and Food-related Outcomes in Children with Autism Spectrum Disorder: Disparities by Income and Food Security

Authors: Anita A. Panjwani^{1,5}, Regan L. Bailey⁵, Bridgette L. Kelleher¹

Introduction: Recent reports on the COVID-19 pandemic have shown the autism community has been considerably affected, especially those who are socioeconomically disadvantaged. However, there is currently a lack of focus on how the changes in food systems and environments are affecting children with neurodevelopmental disorders. Children with autism spectrum disorder (ASD) often follow structured routines, and due to sensory sensitivities, are highly selective in the types of foods they choose.¹ The shortage of certain foods during the pandemic may thus be more challenging for children with ASD, possibly leading to poorer health and nutritional status. However, the impact of COVID-19 on food-related outcomes in ASD has not been explicitly studied. We investigate whether and to what extent children with ASD and their families are being affected by the conditions caused by the global pandemic. The goals of this study are twofold: 1.) to describe the impact of the COVID-19 pandemic on food and eating behaviors of children with ASD and 2.) to understand the potential role of socio-demographic or diet-related factors that may increase risk of negative outcomes, including worsened overall behavior, worsened eating behaviors, and difficulties with food unavailability.

Methods: A total of 200 parents and caregivers across the United States participated in an online survey formulated to understand food environments and eating behaviors in children with ASD. The survey was offered May through June 2020 when most states had COVID-19 regulations in effect. Survey items included questions on respondent and child demographics; socioeconomic status, including food security and availability; food acquisition and preparation methods; child's height, weight, diet, and changes to behaviors, including eating behaviors, prior to and post-COVID-19. Food security was assessed based on a two-question method validated against the US Department of Agriculture 18-item Household Food Security Survey.² This method has been previously used by several studies for assessing household food security status.³⁻⁵ Multivariate logistic modeling was used to model the following outcomes: changes in overall behavior; changes in eating patterns and behaviors; and difficulty in finding foods the child prefers or usually eats. Separate models included either income or food security as dependent variables, as these variables were collinear. While income level was only obtained for prior to the pandemic, food security status was obtained for prior to and post-COVID-19 regulations. Since prior and post-food security status were collinear, these variables were examined in separate models.

Results: A majority (74.4%) of respondents reported a moderate or huge impact on the child's overall behavior, while 57.3% of respondents reported a moderate or huge impact on the child's eating behaviors. Of the respondents who reported to be food secure prior to COVID-19, 22.2% fell into food insecurity post-COVID-19. A household income of \geq \$100K (pre-COVID-19 regulations) was associated with a decreased risk of a moderate/huge impact of COVID-19 on the child's overall behavior

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(adjusted odds ratio (aOR): 0.20, 95% confidence interval (CI): 0.07, 0.58) and on the child's eating behaviors (aOR: 0.31, 95% CI: 0.12, 0.84). Conversely, a household income of \geq \$100K predicted more difficulty with food availability compared to a household income of $<$ \$50K (aOR: 3.79, 95% CI: 1.45, 9.89). Results of food insecurity models paralleled those for household income. However, when compared to *pre*-COVID food insecurity, *post*-COVID-19 household food insecurity status had a greater magnitude of association with the outcomes: overall behavior (aOR: 4.28, 95% CI: 1.89, 9.65), eating behaviors (aOR: 3.01, 95% CI: 1.48, 6.14), and difficulty with food unavailability (aOR: 0.25, 95% CI: 0.12, 0.52).

Discussion: The COVID-19 pandemic and its associated regulations has greatly impacted children with ASD and their families. Our findings show a large proportion of caregivers reporting moderate to huge changes in overall and eating behaviors, which particularly affected those families with low income and/or food insecurity. Conversely, higher income and food security were associated with difficulty in finding the child's preferred foods. This study highlights the cost of existing disparities on children with ASD and their families during this unprecedented time. These findings may inform future policy and interventions to reduce the negative impacts of COVID-19 on individuals with ASD and their families.

References/Citations:

1. Margari L, Marzulli L, Gabellone A, de Giambattista C. Eating and Mealtime Behaviors in Patients with Autism Spectrum Disorder: Current Perspectives. *Neuropsychiatr Dis Treat*. 2020;16:2083-102.
2. Hager ER, Quigg AM, Black MM, Coleman SM, Heeren T, Rose-Jacobs R, et al. Development and validity of a 2-item screen to identify families at risk for food insecurity. *Pediatrics*. 2010;126(1):e26-32.
3. Colizzi M, Sironi E, Antonini F, Ciceri ML, Bovo C, Zoccante L. Psychosocial and Behavioral Impact of COVID-19 in Autism Spectrum Disorder: An Online Parent Survey. *Brain Sci*. 2020;10(6).
4. Sharma SV, Chuang RJ, Rushing M, Naylor B, Ranjit N, Pomeroy M, et al. Social Determinants of Health-Related Needs During COVID-19 Among Low-Income Households With Children. *Prev Chronic Dis*. 2020;17:E119.
5. Tanner K, Case-Smith J, Nahikian-Nelms M, Ratliff-Schaub K, Spees C, Darragh AR. Behavioral and Physiological Factors Associated With Selective Eating in Children With Autism Spectrum Disorder. *Am J Occup Ther*. 2015;69(6):6906180030p1-8.

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Paper Title: Building a virtual treatment network to meet the needs of Angelman families during the COVID-19 pandemic

Authors: Meredith Bucher^{1*}, Eric Shannon^{6*}, Riley Felicicchia¹, Dan Foti¹, Mandy Rispoli⁶, Bridgette Kelleher^{1^}, Kelly LeMaire^{1^}
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Introduction: The onset of COVID-19 has rapidly accelerated virtual treatment to the forefront of clinical care. The advantages of telehealth treatments have been long recognized in rare conditions such as Angelman syndrome (AS), as families affected by this low-incidence condition (~1:10,000) are unlikely to have access to local providers well-versed in the AS phenotype, and AS is associated with sensorimotor impairments, challenging behaviors, and seizures that make travel to specialty clinics difficult. Even prior to COVID-19, caregivers of individuals with AS experienced high unmet needs themselves, including lack of support; limited self-care; chronic, disrupted sleep; and difficulty accessing and managing high-quality childcare and respite care (e.g., Thomson, Glasson, Roberts, & Bittles, 2017). However given the time and effort required to coordinate services for individuals with AS, caregivers' own mental health treatment often takes a back seat, despite substantial evidence that treating caregivers can produce spillover effects that simultaneously benefit their dependents, and vice versa. Here, we present initial data from a Project Well-CAST (Well-being of Caregivers of individuals with Angelman Syndrome through Telehealth), a novel virtual treatment network we deployed during the COVID-19 pandemic to provide both mental health treatment and parent-mediated behavioral coaching to AS caregivers and their young children via telehealth. Treatment was facilitated by an interdisciplinary team of clinical and special education graduate student clinicians, 6 of 8 who were naïve to the AS phenotype. Clinicians

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completed a virtual pre-treatment training on AS, facilitated in part by AS caregivers, and deployed ~10-12 weeks of evidenced based interventions under the supervision licensed professionals. The goal of the present study is to disseminate this novel model for partnering with training clinics to deliver evidence-based treatments virtually during COVID-19, as well as to report initial efficacy and satisfaction data from caregivers enrolled in this trial.

Methods: Twenty-six caregivers (88% female, 12% male; 81% Caucasian, 12% Asian or Pacific Islander, 4% Hispanic, 4% Alaskan) of children with AS were randomized to one of three evidence-based telehealth treatment programs: n=18 received Acceptance and Commitment Therapy (ACT), a cognitive-behavioral therapy that focuses on ways to decrease physical and emotional pain while creating a full, meaningful life (Harris, 2009); n=4 received Enhanced Milieu Training (EMT, Hancock & Kaiser, 2002), an early communication intervention approach that is designed to be embedded into daily home routines; and n=4 received both treatments simultaneously. All caregivers in the ACT treatment groups completed treatment with individual therapists using a set program of weekly topics, exercises, and home practices. Caregivers receiving the EMT treatment watched four online modules and attended weekly coaching sessions targeted to encourage child communication in the natural environment, customized to the child's individual skill profile. Within 24 hours of each sessions, caregivers were asked to complete a series of self-report measures that included the Depression, Anxiety and Stress scale (DASS-21; Lovibond and Lovibond, 1995). They also completed post-treatment measures related to overall telehealth satisfaction. Among participants who engaged in ACT (n=18 total) we used descriptive statistics, *t*-tests, and effect sizes were used to examine treatment effectiveness and overall symptom change among caregivers. Among those who received EMT (n=7), two multiple baseline designs across participants were utilized to assess the effect of the parent training program on the implementation fidelity of caregivers.

Results: Here, we report preliminary findings from 14 caregivers who had completed treatment at time of submission (8 ACT, 3 ACT+EMT, 3 EMT); final analyses will include the full sample and compare outcomes across conditions. Among caregivers engaged in any ACT treatment, results indicated significant differences between self-reported DASS-21 symptoms at Week 1 and Week 9, $t(12) = 2.42, p = .03, d = .33$. Caregivers specifically reported lower levels of depression at the end of treatment compared to the outset $t(14) = 2.83, p = .01, d = .54$. Overall, caregivers reported high satisfaction with the overall experience as well as the ease of technology use, with 82% of caregivers reporting they preferred telehealth services to in-person and 91% stating they would recommend ACT to caregivers of children with AS Caregivers who received EMT increased their use of strategies to enhance their child's communication, demonstrating acquisition criteria (at least 85% fidelity across three consecutive sessions) within 3 to 5 weeks of coaching.

Discussion: Despite the increased popularity of telehealth treatment, particularly during COVID-19, the field lacks models for deploying evidence-based treatments that simultaneously address the needs of caregivers and their children. Preliminary data from project Well-CAST suggest that delivering integrated, specialized treatment for AS families via telehealth is feasible, including generalist training programs in which most trainees have not previously worked with AS patients. The treatments used in this study also appear to be effective, with caregivers reporting high satisfaction and demonstrating clinical gains, both in mental health symptoms and behavioral strategies for supporting their children's development. These findings suggest that telehealth treatments can be an effective model for decreasing barriers and increasing accessibility to a population vulnerable to high levels of stress, including during COVID-19.

References/Citations:

1. Hancock, T. B., & Kaiser, A. P. (2002). The effects of trainer-implemented enhanced milieu teaching on the social communication of children with autism. *Topics in Early Childhood Special Education, 22*(1), 39–54.
doi:10.1177/027112140202200104
2. Harris, R. (2009). *ACT made simple: An easy-to-read primer on acceptance and commitment therapy*. Oakland, CA: New Harbinger Publications.
3. Lovibond, S. H. and Lovibond, P. F. (1995) *Manual for the depression anxiety stress scales*. Sydney: Psychology Foundation.
4. Thomson, A., Glasson, E., Roberts, P., & Bittles, A. (2017). "Over time it just becomes easier...": parents of people with Angelman syndrome and Prader–Willi syndrome speak about their carer role. *Disability and Rehabilitation, 39*, 763-770.