**Symposium Title**: **Engaging Voices: Integrating Stakeholder Perspectives Throughout the Research Process**

**Chair**: Sungeun Kang1

**Discussant**: Rebecca Shaffer 2, 3

**Overview**: Involving key stakeholders, like caregivers, in outcome measures and intervention design is essential for developing clinically meaningful and inclusive outcomes as well as treatment approaches by reflecting the needs and lived experiences of the populations being studied and empowering those directly impacted. During this symposium, presenters will explore how to develop measurement tools that capture the unique insights and experiences of caregivers of and self-advocates with fragile X syndrome (Paper 1), and review the user experience of pre-existing outcome measurement tools, which can significantly impact both research and clinical practices (Paper 2). Additionally, we will discuss how caregiver experiences can help shape more feasible and relevant interventions that address the diverse needs of autistic youth (Paper 3).

**Paper 1 of 3**

**Paper Title**: **Voices of Experience: Caregivers and Self-Advocates on Behavioral Inflexibility in Fragile X Syndrome**

**Authors**: Angelina Jones4, Sungeun Kang1, Rebecca C. Shaffer2, 3, Craig A. Erickson2, 3, Lauren M. Schmitt2, 3

**Introduction**: Behavioral difficulties in individuals with fragile X Syndrome (FXS) are one of the primary reasons families seek medical and psychological support (Weber et al., 2019). Among these, behavioral inflexibility is very common, and when left untreated, can negatively impact quality of life for the individuals with FXS and their families (Bailey Jr et al., 2012). Despite their valuable perspective, caregiver experiences are often underrepresented and overlooked in the creation of standardized research and clinical assessment tools. Caregivers offer critical insights into the everyday challenges and adaptive behaviors of those they care for, yet these experiences are rarely integrated into the development of such tools. This paper addresses this gap by conducting focus groups with caregivers, leading to the development of a caregiver-informed semi-structured interview designed to assess behavioral inflexibility across the lifespan in FXS. The goal was to ensure that caregiver and self-advocate insights shaped both the content and structure of the measure, resulting in a more comprehensive and meaningful assessment.

**Method**: The first phase involved semi-structured virtual focus groups with caregivers of individuals with FXS and self-advocates to gather key stakeholders’ perspectives on their observations of behavioral inflexibility and its impact on daily functioning. Audio-recordings from focus groups were transcribed using NVivo, then verified and coded. Two clinical psychologists reviewed codes to extract primary themes and initially reached >85% agreement across themes and subthemes. Differences were reconciled through discussion and additional review of the transcripts.

**Results**: The analyses revealed several dimensions of behavioral inflexibility that were uniquely emphasized by caregivers. Six primary themes were extracted from transcripts: 1) Intolerance to change, 2) Intolerance to uncertainty, 3) Repetitive interests and behaviors, 4) Family impact, 5) Change in behavior across the lifespan, and 6) Impact of the COVID-19 Pandemic. Our findings showed common examples of these themes including intolerance to disruption to routine, preservative questioning, watching the same things over and over and caregivers having to extensively pre-plan for events.

**Discussion**: The purpose of this study was to gain key stakeholders’ perspectives via focus groups to elicit information and understand patterns of inflexible behaviors in FXS, with the goal of developing caregiver and self-advocate informed disorder-specific measures to accurately assess behavioral inflexibility across the lifespan and in response to treatment. We were able to capture several phenotypic examples of behavioral inflexibility in FXS as well as their impact on individuals with FXS and their families. The findings highlight the importance of including caregiver voices in the development of measures and the need for more inclusive and representative research practices in neurodevelopmental disorders. Findings from the focus groups have subsequently informed the creation of RIGID-FX, or Ratings of Inflexibility in Genetic Disorders associated with Intellectual Disability – Fragile X Syndrome, which is currently being collected in a validation study for which we are also collecting feedback on the feasibility and acceptability of the measure.

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**Paper 2 of 3**

**Paper Title**: Voices of Change: Key Stakeholder Feedback to Improve Caregiver-Report Measures in FXS

**Authors**: Sungeun Kang1, Angelina Jones 4, Rebecca C. Shaffer 2, 3, Craig A. Erickson2, 3, Lauren M. Schmitt2, 3

**Introduction**: Stakeholders are frequently involved in focus groups to help establish content validity when developing outcome measures, but their perspectives on the user experience of completing caregiver- or self-report measures are rarely studied, especially in rare genetic disorders. These insights are crucial for improving the design and implementation of outcome measures reliant on caregiver-report. Available **caregiver-report** measures about behavior are often lengthy, and caregivers are usually asked to complete several assessments covering different behavioral domains, which places a heavy burden on them. This adds to the well-documented caregiver burden and high levels of stress experienced by caregivers of children with developmental disabilities (Masefield et al., 2020). Therefore, this study aimed to gather a range of stakeholder experiences with existing measures through focus groups that included self-advocates with fragile x syndrome (FXS), caregivers of individuals with FXS, and professionals working with this population. Particularly in fragile X syndrome, most respondents of these outcome measures are either caregivers who are premutation carriers (PMCs) of the FMR1 gene or individuals with FXS themselves, it is essential to account for their specific challenges and perspectives.

**Method**: Semi-structured focus groups were conducted with 22 caregivers, 3 self-advocates and 1 professional. All interviews were audio-recorded and transcribed using NVivo12 (QSR International Pty Ltd., 2020). A thematic analysis was used to identify major themes and subthemes. Participants provided suggestions on questions formats and contents, inclusion of certain items, and shared their challenges completing common measures. Initial codes were identified through open coding and finalized through an iterative process by two independent researchers. To ensure reliability, both researchers reviewed the transcripts, identified common themes, and resolved discrepancies through discussion.

**Results**: Thematic analysis identified four overarching themes related to key stakeholders' experiences and perceptions of the caregiver-report measures: Content of the Measure, Structure of the Measure, Potential Accommodations for Completing the Measure, and Impact of the Measure on Caregiver/Self. Each theme was further broken down into a series of sub-themes. Our findings highlighted subthemes and common examples for those subthemes. For example, within the theme of Content of Measure, four subthemes emerged: 1) Encompassing FXS-specific behaviors and 2) Age- and developmentally-appropriate items, 3) Strength-focused items, 4) Items related to the impact of behavior(s) on the quality of life of the individuals and their families.

**Discussion**: This study highlights the importance of including stakeholders' experiences when developing and using caregiver-reports as outcome measures of behavior. Our thematic analysis showed that both the content and structure of the measure are important factors to consider when selecting or designing a caregiver-/self-report measure. Second, we offer crucial insight into the challenges faced by caregivers of people with developmental disabilities while completing report measures as well as potential accommodation that can improve respondent experience during research or clinical visits. We strongly believe that the knowledge gained from our study can be useful for developing improved outcome measures for professionals not only in the FXS field but also for other intellectual and developmental disability fields.

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**Paper 3 of 3**

**Paper Title**: Regulating Together: An Exploration of the Caregiver Experience in a Caregiver-Assisted Group Intervention

**Authors**: Jenna Ruberg 2 , Sungeun Kang 1, Shivali Sarawgi 2, Karen W. Burkett2 , Lauren M. Schmitt 2, 3, Jennifer Harms 2, Rebecca Shaffer2, 3

**Introduction**: Regulating Together (RT) is an evidence-based group therapy program for children experiencing emotion dysregulation (Shaffer et al., 2022). Groups of up to 6 children participate in 10 group therapy sessions while caregivers attend their own group. This novel type of intervention is crucial for those with emotion dysregulation and autism spectrum disorder as autistic children have been observed to exhibit significantly higher levels of emotion dysregulation (ED) compared to their peers in the general population (Conner et al., 2021). Although the clinical importance of ED in ASD has been emphasized (Geller, 2005; Samson et al., 2014), there have been limited studies investigating the efficacy of intervening on ED in autistic populations. RT targets this treatment gap for both children and their caregivers as involving caregivers has been long recognized as crucial for the effective treatment of children with ASD (Brian et al., 2016; Hong et al., 2018). RT stands out because it provides the caregivers with education on the same topics taught in the child group and coaching tips to help facilitate implementation. Due to the essential role of caregivers in RT, and with ASD treatment in general, it is crucial to better understand the perspectives and experiences of caregivers. By exploring the caregivers’ insights into barriers and facilitators to treatment, intervention can be more collaborative and robust.

**Method**: Following the conclusion of the first two rounds of the intervention phase in our RCT using RT (NCT05803369), we asked caregivers to join a 1-on-1 semi-structured interview to share their experience with RT. Trained RT clinicians conducted interviews with 11 caregivers. The interview consisted of questions regarding the caregivers’ experiences in the program, including perceptions of RT's feasibility and accessibility, applicability of skills, the inclusivity of the intervention for diverse groups, as well as the general programming and structure of group sessions.

All interviews were recorded, and the auto-transcribed data was reviewed by study team coordinators to compare data with the original audio recordings. Subsequently, NVivo12 (QSR International Pty Ltd., 2020) was used for data coding and analysis. In the process of conducting qualitative analysis of semi-structured interviews, recurrent patterns are identified as themes (Braun and Clarke, 2006). Throughout the inductive coding process, initial codes were established through open coding, and the final coding framework underwent validation through an iterative procedure. An earlier iteration of themes was presented at Gatlinburg 2024 with the finalized themes and patterns now being presented.

**Results**: In the qualitative analysis, four final themes have now been identified: 1) Caregivers Valued Insights and Learnings, 2) Child Engagement, Learning, and Connectedness, 3) Emotion Regulation Skill Application, 4) Cultural Respect and Inclusivity. These universal themes each have emerged patterns strongly supported by illustrative descriptors to uphold the findings and best represent the caregivers’ experience. In the theme of Caregivers Valued Insights and Learnings, recurrent feedback included the acceptability of the unique concurrent caregiver-child group design, the exceeding of expectations, and a safe space to give caregivers a voice. As expressed by a caregiver, “*I liked that we both learned alongside each other*.” In the theme of Child Engagement, Learning, and Connectedness, caregivers repeatedly placed emphasis on youth being involved in a group setting to learn from peers and the power of shared experiences. A strong endorsement from a caregiver was that for their child “*To see other children with similar issues...it was the main thing for him, to feel like, OK, I'm not the only person*.” In the theme of Emotion Regulation Skill Application, caregivers endorsed feeling competent in applying learned skills, coaching their child and other family members in learned skills, and noticing an increased ability to regulate their own emotions. One caregiver highlighted the material being “*distilled down into some simple principles that you can really apply to just everyday life*." Lastly, in the theme of Cultural Respect and Inclusivity, caregivers highlighted that they felt a sense of belonging and security no matter what culture, family structure, religion, and ethnicity/race, as upheld by the caregiver input that “*Everybody’s beliefs or what they valued or what they taught in their home was respected*.”

Across themes, caregivers reported an overwhelming satisfaction for both them and their child. A caregiver shared feeling that “*it was as much for [them] as parents as it was for [their children*].” Caregivers believed the intentional focus on skill application, optimization of child engagement and skill practice in the structure of RT, the inclusive tone and respectful practices fostered throughout, and group cohesiveness increased caregiver participation, which enhanced their foundational knowledge of ER, use of skills, and feelings of support. Caregivers indicated that they and their children exhibited a healthy willingness to attend and engage in group, as well as the applicability and utility of skills upon group completion.

**Discussion**: Regulating Together has been modified based on feedback from families to create an intervention that is accessible and feasible for families. Interviews revealed valuable discussions about caregivers' experiences with RT, demonstrating its ability to elicit high levels of engagement from caregivers and child participants and provide useful skills, as well as maintain cultural inclusivity. Details embedded in the organization and programming have strategically been created to promote these goals, including accommodations offered to families, aiming to enhance their overall commitment and positive experience. This study demonstrates insights that we obtained about facilitators and barriers to treatment access, impact, and sustainability in ASD by exploring caregivers’ experiences.

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1 University of Nebraska-Lincoln

2 Cincinnati Children’s Hospital Medical Center

3 University of Cincinnati, College of Medicine

4 University of Minnesota