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Efficacy of a Remote Play-Based Intervention for Children with Prader-Willi Syndrome

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Abstract:	The current study examines the efficacy of an 8-week pretend play intervention targeting social-cognitive abilities in children with Prader-Willi syndrome (PWS), ages 6-9. PWS is a rare disorder associated with various social, emotional, and cognitive challenges, linked to pretend play impairments, and for which interventions are sparse. Nineteen children were quasi-randomized to receive the intervention or be part of a waitlist control group. Participants who received the intervention (n = 10), demonstrated significant improvements in various components of pretend play, most notably in organization of play, which may generalize to broader social-cognitive gains. These findings provide evidence of the intervention's efficacy in enhancing pretend play skills and related social-cognitive abilities during this critical period of development for children with PWS.

Abstract

The current study examines the efficacy of an 8-week pretend play intervention targeting social-cognitive abilities in children with Prader-Willi syndrome (PWS), ages 6-9. PWS is a rare disorder associated with various social, emotional, and cognitive challenges, linked to pretend play impairments, and for which interventions are sparse. Nineteen children were quasi-randomized to receive the intervention or be part of a waitlist control group. Participants who received the intervention (n = 10), demonstrated significant improvements in various components of pretend play, most notably in organization of play, which may generalize to broader social-cognitive gains. These findings provide evidence of the intervention's efficacy in enhancing pretend play skills and related social-cognitive abilities during this critical period of development for children with PWS.

Keywords: Prader-Willi Syndrome, pretend play, social cognitive behavior, cognitive flexibility, telehealth

Efficacy of the PRETEND [Play-based Remote Enrichment to Enhance Development] Program for Children with Prader-Willi Syndrome

Prader-Willi syndrome (PWS) is a complex genetic neurodevelopmental disorder attributed to alterations in chromosome 15, either by paternal deletion (DEL; 15q11.2-q13), maternal uniparental disomy (mUPD), or more rarely, an imprinting defect (Driscoll et al., 2019; Lionti et al., 2014). In addition to well-reported hallmark characteristic of hyperphagia and associated challenges, children with PWS also have impaired adaptive functioning and deficits in social cognition and emotional ability (Dykens & Kasari, 1997; Dykens et al., 2019; Dimitropoulos et al. 2017). Moreover, in addition to the strong genetic overlap and increased risk for autism spectrum disorder (ASD) seen in the PWS population (Bennett et al., 2017; Veltman et al., 2004), early social-cognitive abilities of children with PWS have been found to be comparable to those with ASD (Dykens et al., 2017). A recent study showed that children with PWS also have significantly lower emotional competencies, including emotional expression, recognition, comprehension, and regulation, compared to their typically developing peers (Famelart et al., 2022). Furthermore, previous research has consistently shown impaired pretend play abilities in children with PWS similar to children with ASD (Dimitropoulos et al., 2019; Bennett et al., 2015; Zyga et al., 2015). Dimitropoulos et al. (2021) found that children with PWS have limited pretend play skills and cognitive flexibility, displaying more repetitive behavior and rigid thought patterns than a typically developing reference sample.

Pretend play is associated with several aspects of child development including cognitive, socioemotional, and adaptive functioning. Positive associations have been noted between children's pretend play skills and emotion regulation (Hoffman & Russ, 2012; Slot et al., 2017) as well as emotion comprehension (Richard et al., 2021). Pretend play skills also appear to have implications for executive functioning processes such as divergent thinking and cognitive flexibility (Thibodeau et al., 2016; Wallace & Russ; 2015). Fehr and Russ (2016) found that organization in play is directly related to overall cognitive flexibility and divergent thinking ability. Pretend play requires children to operate under the implicit rules of a given story stem or scenario that is separate from the real world. Additionally,

switching between different scenarios or transitioning out of a make-believe realm during a play session can lead to frustration from peers. Improvements in such executive functioning processes through pretend play may be attributable to a child's ability to adhere to such constraints within story stems. (Hassinger-Das et al., 2017). Finally, promoting a child's ability to play with others has been shown to foster the development of social skills such as assertiveness, cooperation, and perspective-taking, which are all important components for facilitating peer relationships (Jaggy et al., 2023; Schlesinger et al., 2020). Given these noteworthy advantages, targeting skill-building through pretend play has been studied as an effective mode of intervention for increasing socioemotional and adaptive abilities, decreasing cognitive and behavioral rigidity, and developing appropriate social engagement in both typically developing children and children with developmental disabilities (Doernberg & Dimitropoulos, 2021).

While behavioral interventions commonly used in other developmental disabilities, such as applied behavior analysis (ABA) or social skills training, may be beneficial for improving various domains of functioning for children with PWS, developing programs specifically for use with a rare-disorder population is difficult due to the rare prevalence of the disorder and geographic variability of impacted families. In a recent study, only 23.4% of caregivers of individuals with PWS reported having access to ABA services (Bedard et al., 2023). In general, behavioral interventions necessitate a large commitment from families and are not often feasible due to the burden of time and associated travel costs. Further, there are additional concerns about the effectiveness of such behavioral interventions, with 66.7% of caregivers reporting that these behavioral supports did not adequately meet the unique needs of their individual with PWS (Bedard et al., 2023).

However, many studies have demonstrated that telehealth interventions can serve as efficient mechanisms of behavioral change for children with developmental disabilities while reducing barriers such as cost, location, and limited access to specialty providers (Langkamp et al., 2015; de Nocker & Toolan, 2021; Vismara et al., 2013). Previous research has demonstrated feasibility, acceptability, and preliminary efficacy of the PRETEND program (Play-based Remote Enrichment to Enhance Development), in increasing the pretend play skills of preschool and school-aged children with PWS

(Dimitropoulos et al., 2017). In the first iteration of the school-aged, direct intervention PRETEND program, parents and children participated in 12 bi-weekly sessions lasting 30-45 minutes across six weeks. Interventionists followed a relatively structured procedure, with all children working towards the same intervention goals regardless of child need or strengths. Dimitropoulos et al. (2021) provided preliminary efficacy of this program, after which children showed increased pretend play abilities across domains of imagination, affect variety, thematic frequency, and organization. Pre- and post-intervention comparisons also showed significant increases in both cognitive flexibility and fluency in thought.

This direct intervention PRETEND program has since been refined to increase parent involvement and adjust weekly time commitment in response to participant feedback from the pilot study. First, the dose of the intervention was altered from twelve bi-weekly sessions to eight bi-weekly sessions in the current study. The duration of these sessions was extended from 30-45 minutes to roughly 60 minutes per session. These changes were made in response to parent feedback and request for additional support at each visit. Additionally, four parent-child joint play sessions were added to control for playtime dosage by providing parents structured time to play with their child and implement skills learned in the parent training sessions. These sessions ensured that parents played with their child at a minimum of once every two weeks over the course of the program, with interventionists providing feedback as parents gained additional insight and play facilitation skills. Additional changes were made to the interventionist manual such as allowing for the individualization of goals of the play sessions to meet the needs of each child.

Finally, the current study included the addition of a waitlist control group to better assess the effects of the intervention over time. The present article aims to examine the efficacy of this refined program in fostering pretend play skills and enhancing social cognition from pre- to post-intervention among school-aged children with PWS. Specifically, it was hypothesized that children who participated in the intervention would show increases in their level of cognitive and affective pretend play skills from pre to post intervention and in contrast to the waitlist control group.

Method

Participants

The present study included 21 children with PWS and their parents. Eligibility criteria required the child to be 6-10 years old, have a PWS diagnosis confirmed via genetic testing, be at least minimally verbal (i.e., be able to produce phrase speech), and be able to sit at a table for study visits and sessions (determined via parent report). Additionally, families needed reliable computer and internet access and use English as their primary language. It was also required that families not be concurrently enrolled in a clinical trial as to not confound any potential results. All parents/guardians participating in the study completed an informed consent and children ages 7 and older provided their written assent. Parents/guardians were compensated for their participation in the study. Payments were distributed based on their completion of each stage of the study (i.e., baseline, outcome, etc.). To assess the feasibility of the remote intervention, participants were recruited from locations at least two hours away from the main study site in Cleveland, Ohio (mean distance = 988.75 miles). Families were recruited through newsletters, online postings, and meetings at the state and national level of the Prader-Willi Syndrome Association (PWSA-USA) and the Foundation for Prader-Willi Research (FPWR).

Quasi-randomization was used to ensure an approximate balance of age, sex, and genetic subtype across the two groups, resulting in comparable groups. 10 families were enrolled in the intervention group (INV) and 11 were assigned to the waitlist control group (WC). Of the 21 enrolled families, one family did not fully complete their baseline visit, and one family was lost to follow-up. Therefore, 19 families were included in pre-/post-analyses. Both families who dropped out of the study had been previously assigned to the WC group.

All demographic and parent questionnaire measures were assessed at baseline prior to randomization. Due to constraints from the COVID-19 pandemic, cognitive reports (Kaufman Brief Intelligence Test-II) were only obtained from the first nine participants. The full-scale IQ of these participants, verbal IQ, and nonverbal IQ were consistent with previous reports in this population (Dykens & Roof, 2008; Jaime, Gerk & Stegmann, 2023). Across the full sample, parents reported below-average adaptive functioning (Vineland Adaptive Behavioral Scales-II) across all five domains: communication,

daily living skills, socialization, motor development, and overall adaptive behavior. Parents also reported above-average problem behaviors with higher scores seen across internalizing behaviors, externalizing behaviors, and overall maladaptive behaviors. Parents additionally reported on child social skills and social communicative abilities in addition to levels of their own parenting stress. The INV and WC groups did not significantly differ on any demographic variables at baseline. See Table 1 for complete demographics.

Measures

Vineland Adaptive Behavior Scales, Second Edition (Vineland – II; Sparrow et al., 2005)

The Vineland-II is a survey rating form completed by parents/guardians that assesses a child's current functioning by providing standard scores in five domains: communication, daily living skills, socialization, motor skills, and maladaptive behaviors. The maladaptive behavior domain is divided into two subscales: internalizing and externalizing behaviors.

Social Skills Improvement System Rating Scales (SSIS; Gresham & Elliot, 2008)

This survey is completed by parents/caregivers and assesses social skills, problem behavior, and autism symptomatology. It is appropriate for use in children ages 3-18. The survey yields scores across 12 subdomains: communication, cooperation, assertiveness, responsibility, empathy, engagement, self-control, externalizing, bullying, hyperactivity/inattention, internalizing, and autism symptomatology. Overall standard scores for social skills and problem behaviors are included, as well as behavior levels (below average, average, above average). The SSIS has been shown to be reliable and valid in special populations (Gresham and Elliott, 2008).

Parenting Stress Inventory (PSI-4; Johnson, 2015)

The PSI-4 is a self-report questionnaire consisting of 120 items that assesses the presence of stress within the parent-child dynamic, appropriate for use in children with developmental disorders (Tomanik et al., 2004). Items are divided into three domains, including child, parent, and situational/demographic characteristics. Average t-scores for this measure falls between 40 to 54.

Social Communication Questionnaire (SCQ; Rutter et al., 2003)

The SCQ is a parent-report questionnaire determining the existence of characteristic autistic behaviors. Resulting scores range from 0-39, with higher scores indicating more social communication impairment.

Affect in Play Scale (APS; Russ, 2004; 2014)

The APS is a standardized assessment examining cognitive and affective processes in play in an unstructured play task. It is appropriate for use in children ages 6 to 10 years old (Russ, 2004; 2014). In the task, children are given two puppets, three blocks, and a standardized prompt in which they are told to play any way they would like for five minutes. This play session is then video-recorded and scored on a criterion-based rating scale. Detailed criteria and operational definitions can be found in the manual (Russ, 2004; 2014). The specific variables evaluated measure organization of the storyline, incorporation of imagination in play, comfort via engagement and enjoyment in play, affect frequency in the narrative, and affect variety. The cognitive variables (*organization*, *imagination*, and *comfort*) are scored on a five-point scale, with scores of one indicating the lowest ability in that domain. The affective variables (*affect frequency* and *affect variety*) are scored via count of units of affective expression and categories of affect. Additional APS variables provided more nuanced insights into pretend play abilities, including interpersonal interaction frequency and type (e.g., *interpersonal positive, negative, neutral*), *thematic frequency*, and *transformations*. Frequency of interpersonal affect type is defined as social interactions between multiple characters that may be positive, neutral, or negative in nature. Additionally, *thematic frequency* defined the number of themes, or different storylines within the play, and *transformations* counted how often the child used an object to represent something else.

Procedure

All study procedures were reviewed and approved by the Case Western Reserve University Institutional Review Board (#2015-1240).

Baseline visit

For participants enrolled prior to the COVID-19 pandemic (n=9), baseline visits were completed in person at a location near the participating family's residence. For participants recruited after the onset

of the pandemic (n=12), baseline visits were completed via Zoom teleconferencing services to accommodate for travel restrictions. During baseline visits, the child completed the APS task while a parent completed surveys regarding the child's overall social, communicative, and adaptive functioning (SSIS, SCQ, and Vineland-II), and the parent's stress levels (PSI-4). Prior to the COVID-19 pandemic, the parent assessments were completed by hand on paper. Participants who enrolled after the onset of the pandemic completed the parent assessments electronically via a survey collection software. There was no change in the APS administration procedure due to the COVID-19 pandemic other than the fact that the measure was administered via video teleconferencing rather than in-person. Initial participants (pre-pandemic) also completed a cognitive assessment (KBIT-2). However, later participants were not able to complete this assessment via telehealth, and thus, this measure was not administered. At the conclusion of the baseline visit, parents were provided with an intervention folder containing the PRETEND parent manual and instructions for videoconferencing.

Intervention program

The school-aged version of the PRETEND intervention program is designed for children with PWS, ages 6-10. It was adapted from a play-based intervention that sought to increase emotional expression and imagination in typically developing children (Moore & Russ, 2008). The program consisted of three session types: weekly direct child play sessions with the interventionist, bi-weekly parent education sessions, and parent-child joint play sessions. The program included a total of 8 direct child play sessions, 4 parent sessions, and 4 parent-child joint play sessions. All INV participants received the same dosage of the intervention. Any conflicts (scheduling, technology-based, etc.) were resolved so that all families completed the same number of sessions. Child play sessions were divided into four modules, each consisting of a different topic to improve social cognition. Topics include "Building Emotional Understanding," "Building Emotional Regulation and Problem-Solving Skills," "Increasing Play Complexity," and "Building Flexibility in Play." One interventionist, a graduate-level doctoral student in clinical psychology was trained to reliability and worked from manualized procedures to ensure fidelity to the intervention program.

Each week, an interventionist played through 2-3 stories with the child during a 15-20 minute play session via videoconferencing software. Previous studies have shown telehealth to be a feasible mode of intervention delivery in this population (Dimitropoulos et al., 2017). Prior to the remote study visit, children were provided a set of toys including dolls, Legos, blocks, toy food, and toy animals, either at the end of the in-person baseline visit or mailed directly to their home. Interventionists had a similar set of toys to allow for modeling and joint play. Interventionists began each story by providing a story stem, a brief statement that set up the story (i.e., "Let's play a story about a girl who goes to the zoo."). These story stems were tailored to elicit specific affective or creative content. Content of the stories varied based on the general interest from each child and module goals, and children were also invited to suggest their own story ideas.

A variety of strategies were used to elicit imagination, organization, and affect during sessions and target specific skills for each of the modules, including modeling, prompting, praise and reinforcement, questioning, labeling feelings, while following the child's lead and encouraging them to produce their own ideas. For example, to show how the blocks might be transformed into something else, the interventionist modeled building and used the blocks as a school. For prompting, the interventionist used phrasing like, "Show me what happens next" to signal to the child to come up with the next plot point and thus practice organizational skills. The interventionist often asked about the emotional state of a certain character, prompting the child to consider how emotions may be at play throughout the story. Praise and reinforcement were commonly used across all modules to increase use of affect, transformations, problem-solving skills, complex plot points, and cognitive flexibility. When modeling emotions in play, the interventionist labeled the feelings of different characters in the story.

In addition to direct play sessions, parents met with their assigned interventionist for four bi-weekly parent education sessions. During these sessions, the interventionist provided background information relevant to that session's topic and reviewed how the information provided in the session could be applied to real-life scenarios. Additionally, parents had the opportunity to review progress made from the previous sessions related to their child's behavior and discuss any challenges they were facing

when implementing the strategies. Four parent-child play sessions were also interspersed throughout the intervention. During these play sessions, parents had the opportunity to put into practice the material they learned in the parent education sessions. Interventionists recorded and reviewed these videos and later discussed effective play facilitation techniques, and their progress on strategies from the parent education sessions. See Table 2 for full description of intervention modules and session goals.

Outcome visit

Parents and children completed an outcome visit within four weeks of their final intervention session. Outcome visits were completed either in a lab setting, a location near the participant, or via Zoom. Children completed the APS again, and parents completed the same surveys from their baseline visit (PSI, SCQ, SSIS, and Vineland-II). As in the baseline visit, families who completed their outcome visit prior to the COVID-19 pandemic completed all assessments in person and, when applicable, via pencil and paper. Families who completed their outcome visit after the onset of the COVID-19 pandemic completed all assessment remotely using video teleconferencing and an online survey collection software.

Results

Two raters independently scored the APS videos for the present study, blinded to group and timepoint. The primary rater was a graduate student in clinical psychology, and the reliability rater was an advanced undergraduate research assistant. Raters completed APS reliability training and were required to reach interrater agreement of .80 or greater before beginning to code for the study. Twenty-five percent of the data was then randomly selected for interrater reliability across both baseline and outcome assessments. Interrater reliability was assessed via intraclass correlations to measure absolute agreement between raters (Hoffmann & Russ, 2012). A two-way random effects model assessed for absolute agreement using a 95% confidence interval. All reliability videos reached interrater agreement of .80 or greater.

Play variables were first tested for significance of changes between groups (INV, WC) across the two study timepoints (baseline, outcome) using mixed factorial ANOVAs. For individual, descriptive analyses, mean APS scores were compared between INV and WC groups. In addition, children in the

INV group who responded to the intervention (responders) were then descriptively compared to non-responders to identify potential distinguishing factors between groups. Response to the intervention was defined by an increase in at least one point on at least one of four core APS variables: *imagination*, *organization*, *affect frequency*, or *affect variety*. Throughout all analyses, participant APS scores were compared to a typically developing reference sample of first and second graders to compare to normative play levels expected for their developmental level (Russ, Robbins, & Christiano, 1999).

Outcome Measures

Affect in Play Scale (APS; Russ, 2004, 2014)

At baseline, APS scores of the PWS sample fell within expected ranges, lower than the typically developing reference group and similar to those seen in previous studies of school-aged children with PWS (Dimitropoulos et al., 2021; Zyga et al., 2015). Children in the WC and INV groups did not significantly differ on any baseline APS variables. Differences between groups across study timepoints were assessed using a series of 2x2 [Group (INV, WC) x Time (baseline, outcome)] mixed factorial analyses of variance (ANOVAs). A significant interaction for the *organization* variable between group and time (p = .021,) was observed. The effect size, as measured by eta squared was η_p^2 = .274, indicating a large effect (Cohen & Wolman, 1965). While falling below statistical significance, other APS variables indicated positive patterns of improvements, including a large effect for *thematic frequency* (η_p^2 = .186, CI, p = .065), and medium effects for *affect frequency*, (η_p^2 = .111, CI, p = .100) and *positive interpersonal* interactions (η_p^2 = .162, CI, p = .087). See Table 3 for full details.

Individual, descriptive analyses were also used to characterize the data due to the small sample size. The INV group demonstrated qualitative improvements in mean APS scores greater than those observed in the WC group. At outcome, the INV group demonstrated higher scores on all APS play variables in comparison to the WC group, excluding the *interpersonal negative* and *transformations* variables, which did not differ between groups at outcome.

Within the INV group, descriptive analyses revealed distinct baseline differences emerged between children who responded to the intervention versus those who did not. As noted, response to the intervention was categorized if scores increased by at least one point in at least one of four core APS variables after the intervention. As previous individual descriptive analyses comparing response to interventions utilized a criterion of 0.5 a standard deviation on the APS (Dimitropoulos et al., 2022), a more conservative criteria was applied in the present study. Specifically, increases in at least one point on the APS was determined to represent a noticeable improvement, representing changes in play ability that can be clearly distinguished according to APS scoring criteria. These differences in APS scoring criteria show robust reliability and thus can be utilized to gauge clinically meaningful and detectable differences in quality of pretend play (Russ, 2004, 2014). While statistical comparisons within the INV group of this study would be underpowered, this method of descriptive comparison may identify nuanced patterns and individual-level changes for participants with PWS (Dimitropoulos et al, 2022). According to these criteria, seven children responded to the intervention and three did not. Children who responded well to the intervention started at qualitatively higher mean APS scores across the core APS variables than their non-responder counterparts. Additionally, the INV responder group demonstrated qualitatively higher mean levels of social skills, lower mean levels of ASD-related social communication impairments, and lower parent-reported stress according to the PSI in the child domain at baseline. The groups were relatively similar on parent-reported maladaptive behaviors as measured by the VABS. See Table 4 for full details. Further, children in the responder group improved after the intervention, such that their mean APS scores were either very close to (e.g., *imagination*), or exceeded scores of the TD reference group, seen in APS *organization*, *affect frequency*, and *affect variety* variables. See Table 5 for full details.

Discussion

The current study continues to support the utility of telehealth play-based interventions for children with PWS. Telehealth interventions serve as a useful mode of service delivery for rare disorder populations such as PWS, addressing barriers to specialized care such as geographic location, transportation, and cost. Additionally, there have been increased calls to evaluate the efficacy of

telehealth interventions since the COVID-19 pandemic, especially within the special needs population (de Nocker & Toolan, 2021; Wainer & Ingersoll, 2015). Participants in this study were able to complete all intervention sessions with limited disruptions or technical difficulties, which provides continued evidence that telehealth platforms can serve as a feasible mode of intervention delivery in children with PWS.

Results from the current study importantly further support the efficacy of the PRETEND program in school-aged children with PWS, in particular, due to the addition of a waitlist control condition. Specifically, several affective and cognitive processes in pretend play improved following the intervention, as evidenced by significant increases in organization, which had a significantly large effect, and noteworthy positive patterns of improvement in frequency of affect expression, positive social interactions, and number of themes exhibited in the play, which also showed medium to large effect sizes. Further, children in the INV group showed mean increases in *organization* from previously deficient to normative ranges expected for their developmental ability (Russ et al., 1999). These improvements were not observed in the waitlist control group from baseline to outcome, which provides evidence that the changes seen in the intervention group stem from the effect of the intervention, rather than time. Further testament to the efficacy of the intervention is that children within the INV group who responded well to the intervention demonstrated pretend play levels that were similar to or exceeded those of developmentally matched peers on the core APS variables (Russ et al., 1999). Of note, there were no significant changes observed in ratings of the children's comfort during play, which suggests that the resulting improvements from baseline to outcome cannot be attributed to increased comfort with the play task, toys, or research staff. Prior research has found the APS to be resistant to practice effects and hold sustained test-retest reliability and validity with repeated attempts (Fehr & Russ, 2016; Hoffmann & Russ, 2016). Thus, it is reasonable to conclude that the observed improvements in APS scores from this study were not simply due to practice with the task, but rather from the intervention itself.

These results are consistent with our previous findings (Dimitropoulos et al., 2021) establishing preliminary efficacy of this program. In both studies, *organization* in play showed significant, consistent improvements following the intervention. Patterns of improvements in *thematic frequency* as well as

affect expression were also seen in each study, though across two different variables, with more prominent gains seen in *affect frequency*, rather than *affect variety*, in the present study. Several additional differences emerged between the two studies. For example, Dimitropoulos et al. (2021) found significant increases in children's *imagination* scores. While this result was not observed by the children in the current study, qualitative mean improvements in *imagination* were found in the INV group. It is possible that these changes may be more robust with a larger sample size.

As the primary outcome measure utilized in this study, the APS captures skills that are not only important for the development of pretend play abilities, but are also crucial to social, cognitive, and emotional development (Russ, 2004). In particular, improvements seen in *organization* in play may indicate amelioration of various closely linked cognitive skills. *Organization* is important in optimal play and refers to structuring play narratives, the use of cause-and-effect, and overall integration of play. Prior research has shown that organization in play directly relates to cognitive flexibility, creativity, and divergent thinking, which is defined as the capacity to produce novel ideas and themes (Fehr & Russ, 2016; Guilford, 1968; Hoffmann & Russ, 2012; Kaugars & Russ, 2009). As the preliminary study documented improvements seen across both *organization* and *thematic frequency* scores alongside a standardized measure of divergent thinking and cognitive flexibility, improvements may be seen in these additional domains had a similar measure been included in the present study (Dimitropoulos et al., 2021). While significant gains seen in *organization* and positive patterns of improvements in *thematic frequency* in the present study are promising indicators that related cognitive flexibility skills may benefit from the intervention, future studies can be strengthened through the addition of an explicit divergent thinking measure to confirm these relationships. Improving cognitive flexibility within pretend play may be particularly relevant as a potential mechanism of addressing challenges with cognitive and behavioral rigidity characteristic to the PWS population.

Although falling below statistical significance, positive changes seen in the expression of affect in the play narrative indicates potential improvements in children's ability to conceptualize and integrate emotion in the play. Affective expression as measured by the APS has been shown to relate to emotional

understanding and expression, skills with which children with PWS particularly struggle (Niec & Russ, 2002; Russ & Grossman-McKee, 1990; Russ & Schafer, 2006). These socioemotional skills are especially important during the school-aged years, which marks a critical period for emotional and social growth for children with developmental disorders, in which they may begin to further diverge from peers. Support in these areas may be optimized during this pivotal time for cognitive and socioemotional development.

In addition to generalizability across skill sets, there is evidence to suggest that the presence of and improvement upon pretend play skills in early childhood may have positive long-term effects. Not only have studies found associations between pretend play skills and divergent thinking in childhood, but longitudinal studies have shown that childhood pretend play skills may relate to divergent thinking later in high school and perhaps into adulthood (Lee & Russ, 2018; Russ, 2016). Relatedly, pretend play interventions have demonstrated promise in increasing behavioral regulation and social behavior relevant to building positive peer relationships in typical populations (Goldstein & Lerner, 2018; Hermann, 2017; Jaggy et al., 2023). While these potential effects on behavior and peer relationships were not immediately captured in the present study, this intervention may similarly support social competence during this critical period for peer relationship development in children with PWS. Further follow-up data for the present study may illuminate the potential for sustained and long-term outcomes of the PRETEND program for children who particularly struggle in these crucial social cognitive domains.

Further, individual, descriptive analyses revealed that most participants (70%) in the INV condition responded well to the intervention, as evidenced by 1-point or greater improvements across at least one core APS variable. The children that responded to the intervention demonstrated more complex play skills, greater social skills, and fewer impairments in social communication at baseline assessment compared to those who did not respond to the intervention. Parents of INV responders also reported lower levels of stress at baseline. However, both the children who responded to the intervention and those that did not exhibited similar levels of VABS-2 maladaptive behaviors. These findings may suggest that some children with PWS may benefit from more support prior to engaging in this type of intervention, such as

ensuring they have a strong foundation of social and communicative skills to build from. As such, it may be that pretend play intervention is not as appropriate for children with PWS with greater deficits, requiring a pre-existing level of social and play abilities in order to engage with an interventionist. Additionally, it may be possible that parents who participate in similar programs benefit more from the intervention when not overburdened by stress related to caring for their child. Future investigation into intervention readiness, fit, and responses across different profiles of children with PWS may better elucidate this phenomenon (Dimitropoulos et al., 2022). Further, assessment of potential intervention readiness and adjustment for these factors can ensure that similar interventions maximize family benefits through increased sensitivity to participant needs and abilities.

There were several limitations in the current study. Due to the rarity of PWS, the sample size was relatively small, with a total of 19 families completing the study. As a result, it was not possible to assess for treatment-related differences across genetic subtypes of PWS. In recruiting participants, there were no exclusion criteria regarding past or concurrent services, except for the involvement in another clinical trial. Thus, children could have received additional services over the course of their participation in the study, potentially confounding the results. While the dosage of parent-child playtime was somewhat controlled for through parent-child play sessions, it was not possible to control for additional parent-child play that occurred outside of the scheduled intervention sessions. Therefore, the dosage of parent-child play practice likely varied from family to family. Additionally, given the remote nature of the study and the COVID-19 pandemic, in-person assessments could not be collected, such as measures of divergent thinking and cognitive flexibility, which research has found to be related to overall levels of creativity and related pretend play skills. As such, the present study was unable to explore any direct effects of the intervention on cognitive flexibility or divergent thinking. Further, a robust literature has demonstrated linkages between the APS and other social, cognitive, and emotional skills (Fehr & Russ, 2016; Guilford, 1968; Hoffmann & Russ, 2012; Kaugars & Russ, 2009l Niec & Russ, 2002; Russ & Grossman-McKee, 1990; Russ & Schafer, 2006;). However, the present study only examined these abilities within the pretend play context, which functions as a measure and mechanism for broader social cognitive skill

growth. Additional, explicit measures of these socioemotional skills that closely relate to the APS may more directly illuminate the hypothesized cascading impacts of pretend play skill development for this population.

Future research in this area may benefit from several considerations, including the utilization of cognitive flexibility, divergent thinking, and other social cognitive measures when available. Additionally, this study should be replicated in larger sample sizes to increase its potential for generalizability. As noted, further examination of the long-term impact that pretend play interventions have for children with developmental disabilities may underscore the significant potential of the PRETEND program in supporting pretend play and related social skills for children with PWS.

Overall, the current study provides further evidence for the efficacy of a direct, play-based remote intervention for school-aged children with PWS. In addition to supporting preliminary findings from this program, this work contributes novel information toward intervention efforts for children with PWS. Specifically, remote pretend play interventions continue to serve as a feasible and efficacious mode of improving play skills in this population, with the potential to strengthen associated cognitive and socioemotional abilities in middle childhood. Despite the small sample size, participants improved significantly in their organization in play, and promising patterns of improvements were found across several other cognitive and affective components of pretend play. Additionally, this study provides evidence for the use of pretend play as an effective, enjoyable, and accessible mode of intervention delivery within a developmental disabilities population. Remote, play-based interventions like the PRETEND program have the potential to support families and children with neurodevelopmental disabilities during this critical period of child development, building a foundation for more robust, impactful social cognitive skill growth.

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Table 1
Baseline Participant Descriptives

M (SD)	Total (n = 19)	INV (n = 10)	WC (n = 9)
Age	7.68 (1.00)	6.24 (1.14)	7.68 (0.89)
Gender (% male)	8 (42.10)	5 (50.00)	3 (33.33)
Genetic Subtype (% DEL)	5 (26.31)	7 (70.00)	5 (55.56)
Race (% Non-White)	3 (15.79)	1 (10.00)	2 (22.22)
Verbal IQ [a*]	70.88 (13.37)	73 (12.90)	73 (n/a)
Nonverbal IQ [a*]	67.00 (14.06)	66.57 (15.13)	48 (n/a)
Full Scale IQ (FSIQ) [a*]	64.75 (13.09)	65.71 (13.83)	55 (n/a)
Child Domain Stress [b+]	55.26 (8.11)	54.20 (9.15)	56.44 (7.14)
Parent Domain Stress [b+]	51.21 (10.84)	50.20 (9.40)	52.33 (12.54)
Total Stress [b+]	53.16 (9.19)	52.10 (5.12)	54.33 (9.96)
Life Stress [b+]	44.11 (5.54)	43.90 (6.52)	44.33 (4.58)
SCQ [c+]	9.68 (5.98)	8.70 (6.62)	10.78 (5.36)
SSIS Social Skills [d]	80.89 (13.36)	81.20 (16.73)	80.56 (9.32)
SSIS Problem Behaviors [d]	106.84 (10.67))	106.90 (12.05)	106.78 (9.62)
Vineland Adaptive Behavior Scales			
Communication	82.42 (9.04)	83.20 (10.68)	81.56 (7.35)
Daily Living	81.32 (11.79)	79.10 (6.90)	83.78 (15.86)
Socialization	82.28 (11.28)	84.78 (11.23)	79.78 (11.40)
Motor Development	70.17 (5.46)	72.67 (4.62)	67.67 (5.86)
Adaptive Behavior Composite	78.56 (7.06)	79.44 (8.09)	77.67 (6.22)

[a] KBIT-2: Kaufman Brief Intelligence Test
[b] PSI: Parenting Stress Index (Standard Score)
[c] SCQ: Social Communication Questionnaire (Total Score)
[d] SSIS: Social Skills Improvement System (Standard Score)
[*] n = 8
[+] Higher scores = greater impairment

Table 2
PRETEND Intervention Module Sessions, Themes, and Goals

Week	Session Type	Theme	Child Play Goals
1	Session 1: Child Play Session	Building Emotion Understanding	Introduce emotions in play. Help the child to label, express, and integrate positive and negative emotions.
	Session 2: Parent Education	How to Be a Play Partner	
2	Session 3: Child Play Session	Building Emotional Understanding	
	Session 4: Joint Play Session [a]	N/A	
3	Session 5: Child Play Session	Building Emotion Regulation	Work through problems the child faces and ways to respond appropriately. Model and play through coping tools and problem-solving techniques.
	Session 6: Parent Education	Building Emotions and Complexity in Play	
4	Session 7: Child Play Session	Building Emotion Regulation	
	Session 8: Joint Play Session [a]	N/A	
5	Session 9: Child Play Session	Increasing Play Complexity	Increase imagination to include novel pretend events outside of daily experience, such as going to the moon or having superpowers.
	Session 10: Parent Education	Understanding Your Child's Behavior	
6	Session 11: Child Play Session	Increasing Play Complexity	
	Session 12: Joint Play Session [a]	N/A	
7	Session 13: Child Play Session	Building Flexibility in Play	Emulate play of a peer, changing endings, characters, or themes the child typically gravitates toward. Model and play through novel events and twists and turns in play.
	Session 14: Parent Education	Increasing Social Engagement	
8	Session 15: Child Play Session	Building Flexibility in Play	
	Session 16: Joint Play Session [a]	N/A	

[a] Joint Play Sessions occur between parent and child, in person and recorded via video for interventionist review.

Table 3
Mixed Factorial ANOVA results for INV vs. WC (Whole Sample) from Baseline to Outcome

APS Variable M (SD)	TD Reference Group [a] (n = 30)	INV Baseline (n = 10)	INV Outcome (n = 10)	WC Baseline (n = 9)	WC Outcome (n = 9)	*p*-value	Effect Size η_p^2
Imagination	2.80 (1.13)	1.80 (0.63)	2.10 (0.99)	2.00 (1.12)	1.78 (0.67)	.188	.100
Organization	2.90 (1.21)	1.50 (0.71)	2.80 (1.75)	1.67 (1.12)	1.56 (0.73)	.021*	.274
Comfort	3.30 (1.12)	3.60 (1.17)	3.20 (1.14)	3.11 (1.27)	3.11 (1.05)	.145	.121
Affect Frequency	14.30 (12.64)	3.90 (6.47)	10.30 (10.67)	6.67 (9.51)	4.44 (5.79)	.100	.151
Affect Variety	3.10 (1.86)	1.60 (1.78)	2.70 (2.58)	1.56 (1.88)	1.44 (0.88)	.164	.111
Thematic Frequency	--	1.60 (1.35)	2.60 (2.01)	2.78 (2.95)	2.22 (1.48)	.065	.186
Transformations	--	0.80 (1.03)	1.00 (1.70)	1.67 (2.50)	1.11 (1.05)	.415	.039
Interpersonal Positive	--	0.70 (1.34)	1.70 (3.09)	0.89 (2.32)	0.22 (0.44)	.087	.162
Interpersonal Negative	--	0.00 (0.00)	0.20 (0.63)	2.78 (7.97)	1.56 (4.67)	.650	.012
Interpersonal Neutral	--	2.00 (275)	5.80 (9.35)	1.44 (1.67)	1.22 (1.09)	.214	.089

* = $p < .05$
[a] Russ, Robbins, & Christiano, 1993

Table 4
Baseline Characteristics between INV Responders vs. Non-Responders

M (SD)	Responder (n = 7)	Non-responder (n = 3)
Age	7.89 (1.21)	7.24 (0.98)
SCQ [a+]	7.86 (6.41)	10.67 (8.08)
SSIS Social Skills [b]	85.29 (16.85)	71.67 (14.57)
SSIS Problem Behaviors [b]	105.14 (912)	111.00 (19.08)
PSI-4 Child Domain [c]	51.29 (9.25)	61.00 (4.58)
VABS Adaptive Behaviors [d]	80.57 (8.87)	75.50 (3.53)
VABS Maladaptive Behaviors [d]	18.49 (2.27)	19.00 (2.00)

[a] SCQ: Social Communication Questionnaire (Total Score)
[b] PSI: Parenting Stress Index (Standard Score)
[c] SSIS: Social Skills Improvement System (Standard Score)
[d] VABS: Vineland Adaptive Behavior Scale-II (Standard Score)
[+] Higher scores = greater impairment

Table 5
Baseline and Outcome APS Scores for INV Responders vs. Non-Responders

APS Variable M (SD)	Responder Baseline (n = 7)	Responder Outcome (n = 7)	Non-responder Baseline (n = 3)	Non-responder Outcome (n = 3)	TD Reference Group [a] (n = 30)
Imagination	2.00 (0.58)	2.43 (0.98)	1.33 (0.58)	1.33 (0.58)	2.80 (1.13)
Organization	1.71 (0.76)	3.57 (1.51)	1.00 (0.00)	1.00 (0.00)	2.90 (1.21)
Comfort	2.71 (1.25)	3.57 (0.98)	2.33 (1.15)	2.33 (1.15)	3.30 (1.29)
Affect Frequency	5.42 (7.32)	14.71 (9.74)	0.33 (0.58)	0.00 (0.00)	14.30 (12.64)
Affect Variety	2.14 (1.86)	3.86 (2.19)	0.33 (0.58)	0.00 (0.00)	3.10 (1.86)
Thematic Frequency	2.00 (1.41)	3.43 (1.81)	0.67 (0.58)	0.67 (0.58)	--
Transformations	1.14 (1.07)	1.43 (1.90)	0.00 (0.00)	0.00 (0.00)	--
Interpersonal Positive	1.00 (1.53)	2.43 (3.51)	0.00 (0.00)	0.00 (0.00)	--
Interpersonal Negative	0.00 (0.00)	0.29 (0.76)	0.00 (0.00)	0.00 (0.00)	--
Interpersonal Neutral	2.71 (3.04)	8.29 (10.36)	0.33 (0.58)	0.00 (0.00)	--

[a] Russ, Robbins, & Christiano, 1993