

# Evaluating the Feasibility of a Play-Based Telehealth Intervention Program for Children with Prader–Willi Syndrome

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**Abstract** Here we report the feasibility and acceptability of telehealth for direct intervention in children with Prader–Willi syndrome (PWS). Children with PWS have social-cognitive challenges that are similar to children with ASD. However, developing behavioral interventions for individuals with PWS is faced with the significant challenge of enrolling enough participants for local studies where multiple visits per week are indicated for effective intervention. This study delivered a 6-week play-based intervention via telehealth *directly* to eight children with PWS (6–12 years). Participants completed the program with minimal behavioral or technological difficulty (#sessions  $M=11.875/12$ ). Behavioral Intervention Rating Scale results indicate good acceptability ( $M=5.54/6.00$ ). These findings support using telehealth in rare disorders and delivering intervention directly to children with developmental delays through this modality.

**Keywords** Prader–Willi syndrome · Telehealth · Play-based intervention · Video conferencing

## Introduction

Prader–Willi syndrome (PWS) is a congenital genetic neurodevelopmental disorder that is characterized by intellectual impairments, hyperphagia, intense food preoccupations, obesity, characteristic appearance, and maladaptive

behavior (Cassidy et al. 2012; Dykens et al. 1999). PWS affects 1 in 12,000–15,000 individuals and is caused by an absence of paternally active gene expression on the proximal arm of chromosome 15(q11–q13) (Cassidy et al. 2012). Individuals with PWS experience impaired adaptive functioning, due not only to intense food preoccupation and problem behaviors that stem from difficulty with regulating nutritional intake, but also because this population shows decreased social and emotional functioning as compared to typical individuals (Dykens and Kasari 1997; Dykens and Rosner 1999; Holland et al. 2003). Specific impairments exhibited by individuals with PWS may reflect difficulty in interpreting and using social information and regulating emotions effectively. Further, individuals with PWS express patterns of repetitive behaviors, deficits in social responsivity and competence, and high externalizing behaviors surrounding rigidity (Descheemaeker et al. 2006; Dimitropoulos et al. 2013; Dykens et al. 2011).

Preliminary research has also found that children with PWS have impaired pretend play abilities that are similar to children with autism spectrum disorder (ASD) (Bennett et al. 2015; Zyga et al. 2015). This decreased pretend play ability seen throughout childhood in PWS may be a marker of larger social cognitive deficits characteristic of the disorder. Further, given that pretend play is related to a host of positive outcomes, relating to social awareness (Bergen 2002; Lillard 1993), emotion regulation (Hoffmann and Russ 2012; Moore and Russ 2006), and even language development (Bergen 2002; Charman et al. 2000), targeting skill building through play in children with PWS may strengthen adaptive skills, socioemotional understanding, and the ability to appropriately interact in social situations while also decreasing rigid and repetitive behaviors.

However, developing behavioral interventions for individuals with PWS is faced with the significant challenge of

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enrolling enough participants for local studies where multiple visits per week are indicated for effective intervention. Successful behavioral intervention requires multiple interactions between clinicians, families, and clients, something that is nearly impossible to implement when families live more than 30 miles from the clinic. And for a rare disorder such as PWS, it is common for families to travel regionally and sometimes nationally for research participation and clinic visits. Further, developing efficacious interventions tailored for a specific population requires large sample sizes. These types of studies are needed in PWS and finding innovative ways to provide intervention is essential.

Telehealth methodology is changing the face of intervention today and may reduce the barriers to developing interventions for individuals with PWS. Remote video conferencing as a treatment modality has been shown to be successfully used with rare disorders such as sickle cell disease, cystic fibrosis, and complex pain syndromes (Cox et al. 2012; Jennett et al. 2003; McGeary et al. 2012). This can involve live video conferencing from the home where participants and their families can interact with care professionals and receive support at a distance. Telehealth is gaining a larger presence in the disabilities community, both in research and clinical practice for assessment (Harrell et al. 2014; Parmanto et al. 2013), parent training (Meadan and Daczewitz 2015; Wainer and Ingersoll 2015), and direct intervention (Duncan et al. 2014) for child and adult populations. In adult populations, Parmanto et al. (2013) has developed an integrated telehealth system to screen for ASD. Studies have also investigated the use of telehealth in helping aging individuals with developmental disabilities gain more independence and check in with providers remotely (Agree 2014; Brewer et al. 2010). A growing literature has begun to examine the use of telehealth with parents and caregivers of children with developmental disorders. For example, in Fragile X, telehealth has been used to deliver parent-implemented language interventions to young boys with the syndrome, either through video coaching strategies or the use of interactive touchscreen technology (Díez-Juan et al. 2014; McDuffie et al. 2016). In ASD, much intervention work has been conducted in training parents on behavior management and functional communication techniques in improving problem behaviors and overall functioning (Vismara et al. 2013; Wacker et al. 2013).

Given increased use and acceptability, telehealth and related technology-based methods of treatment delivery have the potential to significantly augment or even replace traditional service models. Research suggests that there are numerous benefits associated with the use of telehealth, such as providing cost-effective intervention options, ease of access to services, and possibly increasing provider system coverage relative to traditional in-person service options (Langkamp et al. 2015; Wainer and Ingersoll 2015).

Via telehealth, users are able to interact directly with clinical researchers and instructional content, which gives providers an ability to track progress and patient understanding seamlessly through technological applications, such as active learning tasks and feedback surveys. The benefits of telehealth technology, the ease of access to this type of treatment, and initial evidence suggesting the feasibility and efficacy of delivering assessment, parent training, and even direct intervention to older individuals via this method suggest that telehealth applications may serve as a promising alternative to traditional in-person treatment (Ingersoll et al. 2016; Meadan and Daczewitz 2015).

Based on the current status of telehealth research, an important question moving forward is whether a telehealth intervention could be effectively delivered to a child alone if that child presented with developmental delays or disorder. Specifically, in children with PWS, could the use of play to help increase imagination and flexibility be possible within a telehealth format? Previous work, across both typical and atypical development, has shown that the use of pretend play interventions can lead to increased flexibility, imaginative thought, and ability to engage in play with others (Barton and Wolery 2008; Moore and Russ 2006). In extending this work to children with PWS via telehealth methodology, the current study reports on a pilot study aimed to deliver a 6-week play-based behavioral intervention *directly* to children with PWS, ages 6–12 years, twice a week, for a total of 12 sessions. During these 15–20 min sessions, the interventionist and child interacted individually with one another via distance video conferencing, with no added support from a parent or caregiver. To our knowledge, this is the first report of direct play-based intervention using a remote interface in children with a neurodevelopmental disorder. Given this, the focus of this article is to report on the feasibility of using telehealth for direct intervention in a PWS sample. The efficacy of the intervention, as it relates to standardized behavioral outcome measures, will not be discussed here. Instead, the methodology of telehealth as an intervention will be reported on in determining if it is possible to play with a child over an online format and how parents and children with PWS react to a play-based intervention structure. Findings regarding the intervention's feasibility and acceptability, as measured by parent report, will also be discussed and compared to traditional service models.

## Methods

### Participants

Ten children with PWS (seven males; three females) and their parents participated in the current study. Eligible

participants provided confirmation of a diagnosis of PWS by genetic testing, were between the ages of 6–12 years of age, had reliable internet access in the home, and were able to complete both pre- and post-assessment visits in person. While the majority of children with PWS function in the mild intellectual disability to low average intelligence range of functioning, studies have shown that there is variability across the population with scores ranging from mid-40s to low 90s (Whittington and Holland 2017). In addition, some children with PWS have delayed language development or speech language disorders (Lewis et al. 2002), thus criteria for inclusion included displaying adequate verbal ability to interact with the interventionist and the ability to independently sit at a table to complete assessment tasks and intervention sessions. Cognitive ability as measured by the Kaufman Brief Intelligence Test (Kaufman and Kaufman 2004) indicated a sample mean Full Scale IQ=67.22 (SD=9.81), within the range of what is expected for the PWS population.

Potential participants were recruited from areas that were distant from the main study site (at least 2 h away from Cleveland, OH; mean distance: 844.4 miles) in order to test the feasibility of intervention when face-to-face contact was truly not a possibility during the intervention period. All participants were recruited through newsletters, website postings, and announcements at meetings of surrounding state and national chapters of the Prader–Willi Syndrome Association (PWSA-USA) and the Foundation for Prader–Willi Syndrome Research (FPWR). Out of the ten participants who enrolled into the program, eight completed the intervention and post-intervention assessments. Factors leading to disenrollment included the inability to dedicate time to the intervention program every week and to sit independently at the computer during intervention sessions.

### Settings and Materials

All participants completed the telehealth intervention program using their own home computers and internet connections. To control for video quality and to ensure a widescreen viewing experience, all participants were provided with a webcam (Logitech HD Pro Webcam C920) and instructions for use during the intervention. To ensure standardization across the intervention, during the baseline in-person assessment each participant was also provided with toys and materials, including a parent manual and handouts, needed for the telehealth sessions. Toys included human figures, blocks and Lego pieces, a miniature car, animal figurines, and plastic food items. Participants were instructed to keep the toys and materials safe and to only use them during the scheduled sessions with the interventionist.

All sessions took place in the homes of each participant and were conducted over internet-based, invitation-protected video conferencing software. All sessions were recorded using commercially available screen-recording software and kept in the lab of the PI (Dimitropoulos) for later coding and review. Prior to the first intervention session, the parent, child, and interventionist conducted a technology check session, where the parent ensured that the home computer was able to connect to the video conferencing software and the web camera was compatible with the home computer. The interventionist and parent also strategized about how to position the web camera for optimal viewing of table space and materials needed for each session.

### Study Procedure

#### *Eligibility*

To be scheduled for a baseline visit, families had to meet eligibility criteria (i.e., be a child with PWS between the ages of 6–12, not currently enrolled in any other pharmacological or behavioral study or therapy that may aim to alter behavior, be minimally verbal, and be able to sit at a table or computer for at least 30 min) and also voice both interest and time availability in attending two video conference sessions a week for a 6-week period. Once these families were identified, a study coordinator would reach out to the potential participants about scheduling an in-person baseline assessment at a location close to their home. One week before the baseline visit, the potential participants were sent an itinerary via email, which provided details on the testing location, parking, and what the research visit would entail. The research team would then travel to this location and conduct baseline visits and enroll families into the intervention program.

#### *Baseline Visit*

During the initial baseline visit, interested families met with the researchers and discussed the structure of the intervention program and technological considerations. If the family was still interested and able to provide availability to schedule sessions with the interventionist, they then underwent the consent process and completed baseline assessments. Following assessments, the child was introduced to the intervention materials (i.e., a standardized set of toys) and did a warm-up, 10-min play session with their assigned interventionist using the provided materials. After this warm-up period, the interventionist explained to the child that they would be using the toys again and playing over the computer just as they had done at the warm-up session. If needed, the interventionist would also use a

laboratory computer, which already had the webcam and video conferencing software installed, to demonstrate the online procedures to the child. Upon completion, the parent was given the webcam and an intervention folder, which included a parent manual and instructions for how to connect the webcam to a computer and how to connect to the video conferencing software. Lastly, the parent and interventionist scheduled a technology check session approximately 1 week from the baseline visit. Intervention sessions began 1 week after the technology check (2 weeks after the baseline visit) given no technological issues.

*Intervention Program*

The intervention program used in the current study was adapted from a play-based program aimed at increasing imagination and emotional expression in typically developing children (Moore and Russ 2006). The program includes 12, 15–20 min sessions delivered directly to the child twice a week for 6 weeks. The 12 sessions were broken down into 3 sections (4 sessions each) that focused on a different skill set relating to emotional expression and understanding and to self-coping techniques. Each participant was assigned an interventionist, who conducted all sessions with that participant. Two interventionists were used in the current study and followed manualized procedures and goals for each session to ensure fidelity in targeting the specific skills being taught during the intervention program. The sessions lasted 15–20 min in length, where the interventionist would model play, social interactions, emotional understanding, and emotional regulation skills directly to the child through engaging in a specific story stem (i.e., Let’s make up a story about ...). As the goal for each session changed, so too did the story stems to guide the play and building of specific skill sets. Specifically, sessions 1–4 focused on building emotional recognition and expression. Session 1 included introducing the toys and how to use them during intervention sessions along with working on labeling one emotion (e.g., happy) in a story stem about “going to the zoo.” In session 2, the interventionist would then introduce two or more positive emotions within the play. The goal of this session would be to construct a story using both of the positive emotions. Session 3 introduced negative emotions and the pair would work through a story expressing both positive and negative emotions. Lastly, in session 4, the interventionist and participant continued to work on integrating positive and negative emotions with a goal of expressing at least three emotions during the play period. The next set of sessions (5–8) focused on building emotion regulation skills. In session 5, the interventionist engaged the participant in a story stem that relates to a problem the child usually faces as reported by the parent. The aim was to work through the problem in a play-based manner and

suggest various ways to act differently. Session 6 continued work on this problem behavior by practicing alternative strategies. Session 7 transitioned to working on a more difficult situation or behavior the child faces (i.e., being nervous about playing with others or going to school), and the interventionist worked through various coping techniques with the child. In session 8, the interventionist would ask the child to present a problem or time when they got upset and to play out how they would fix it. The last set of sessions (9–12) focused on teaching skills related to transitioning and flexibility. In session 9, the interventionist focused on changing routines or having to stop a preferred activity and would play through a story stem where a character had difficulty with transitions. Next, in sessions 10 and 11, the interventionist would work through the previously discussed difficulty in transitioning and practice strategies that may help. As a culmination of the intervention program, in session 12, the child was asked to come up with a difficult situation and use the skills they learned (identifying/expressing emotions, coping skills) to play through how to resolve the situation (see Table 1 for full intervention schedule).

Outside of the 12 child-directed sessions, the intervention also included three parent coaching sessions scheduled to occur during weeks 1, 3, and 5 of the intervention period. One parent from each family was asked to participate in all the sessions. Parent coaching sessions were adapted from a standardized manual intended to teach play and engagement skills to children (Moore and Russ 2006). Parents were provided a printed version of the manual to follow along with during these sessions. In

**Table 1** PWS telehealth intervention schedule

Time points	Events
Enrollment	Pre-intervention assessment (in-person)
Week 1	Intervention session 1 Intervention session 2 Parental training session 1
Week 2	Intervention session 3 Intervention session 4
Week 3	Intervention session 5 Intervention session 6 Parental training session 2
Week 4	Intervention session 7 Intervention session 8
Week 5	Intervention session 9 Intervention session 10 Parental training session 3
Week 6	Intervention session 11 Intervention session 12
Study termination	Post-intervention assessment (in-person visit within a month of session completion)

particular, parent coaching session 1 focused on engaging their child in play and interactions, session 2 focused on building emotional understanding, and session 3 focused on understanding different coping strategies to increase emotion regulation.

**Interventionist Training and Fidelity** As mentioned above, two interventionists worked with the participants in the current study. The first interventionist (O. Zyga) was involved in creating the manualized program with the other two authors and went through extensive pre-pilot work to ensure that the program material was being properly delivered to potential participants. Once intervention sessions began, video recorded sessions were reviewed by the interventionist and third author to ensure the manual was being delivered as intended. The second interventionist underwent extensive training in the protocol prior to administering the intervention. Specifically, the second interventionist first reviewed the manual and previous recorded participant sessions. Next, this interventionist practiced sessions via the online format with the first interventionist. These sessions were recorded and reviewed by the first and third author to ensure that the aims of the intervention sessions were being captured and that the delivery of the content was similar to the first interventionist. Once the second interventionist began delivering the program to participants, their recorded sessions were also reviewed by the third author to ensure fidelity.

**PWS Telehealth Program Delivery** During the intervention program, all communication occurred via email to the family's already established account or through phone conversations between the participant, parent, and interventionist. The first video conference session to occur with each family was a technology check scheduled a week after the baseline visit. Before the first tech check session, and all subsequent intervention sessions, a link to the virtual meeting room for that session was sent to the participant with instructions (i.e., "click this link to connect into the intervention session"). The parent was instructed to click into this link at the time of the scheduled session. Once the link was accessed, a separate window opened with a video stream. During the tech check session, the interventionist directed the parent through the webcam setup and ensured that the camera angle allowed for a complete view of the table where the child would be sitting with the intervention toys.

After setting a schedule for twice weekly sessions with the parent and child, the interventionist would send an email with the virtual meeting room link to the participant on the morning of each session, so as to serve as a reminder. If sessions had to be canceled or rescheduled, the parent was asked to email or call as early as possible before the session time. Given the pilot phase of the current study,

no limit was set on the number of sessions that could be cancelled or rescheduled.

During each intervention session, the child was asked to sit at a table in front of the computer with their toys. The interventionist had access to a duplicate set of toys and would guide the child through the story stems that were appropriate for that day's lesson based on the target skill set. For example, if the focus of the session was to build symbolic thought, the story stems may have related to "going to the moon" or "exploring an underwater city." Parents were asked to be nearby (most often in another room) if any technology issues arose, but were instructed not to participate in the sessions in any way. If the child protested or disengaged during the session, the interventionist used a standard format of prompts to re-engage the child. As a last option, the session could be rescheduled if the child was too tired or other factors (too close to meal times; too many distractors) arose.

#### *Post-intervention Visit*

After completion of the 12 intervention sessions, the post-assessment visit was scheduled with each participant. These visits occurred 1–4 weeks after completion of the program (mean number of weeks post-intervention = 2.68; SD = 1.74) and were conducted in person with the participant. During this visit, parents completed the same surveys and the child underwent the same assessments as had occurred at the baseline visit. During this final visit, participants were thanked for their participation and any final questions or concerns were discussed with the research team.

#### **Program Acceptability**

Parents of participants were emailed a link to complete a modified version of the Behavioral Intervention Rating Scale (BIRS; Elliott and Treuting 1991) after the final intervention session and post-intervention visit. Completion of the survey was optional; if participants chose to complete the modified BIRs, they did so anonymously and provided no information regarding themselves or their child on the form. The BIRs has been shown to be a valid and reliable measure of treatment effectiveness and acceptability. Specifically, these factors have been shown to highly correlate with other treatment outcome measures (Martens et al. 1985) and show strong internal reliability and consistency (coefficient alphas of 0.97 for acceptability factor and 0.92 for effectiveness factor; Elliott and Treuting 1991). In this study, the BIRs was modified to better capture the goals of the intervention program (i.e., building social, emotional, and play skills) and the method of service delivery (i.e., online interventionist–child interactions, remote coaching



sessions). This survey prompted parents to report on areas relating to: (1) appropriateness of the content of the intervention, (2) interactions with the interventionist, (3) their experience in setting up and using the technology related to the intervention, and (4) overall satisfaction with participating in the program. In total, the modified BIRs provided information on domains related to intervention acceptability (13 questions), intervention effectiveness (six questions), and intervention usability (six questions). Parents were asked to respond to questions in each of the domains by rating their replies on 6-point Likert scales (1 = strongly disagree; 6 = strongly agree), overall satisfaction on 5-point Likert scales (1 = extremely negative; 5 = extremely positive), and how the telehealth intervention compared to standard treatment options and likelihood parents would recommend this modality of treatment on 4-point Likert scales (1 = extremely different/unlikely; 4 = extremely similar/likely). Lastly, parents were asked to indicate any limitations of the telehealth model and areas for improvement in an open-ended format.

## Results

### Program Feasibility

#### *Technological Considerations*

All families enrolled in the study were able to communicate with research personnel via already existing email addresses. One participant had to create a new email account in order to access the video conferencing software. Further, all participant’s home computers were compatible with the Logitech webcam provided with the study

materials. The most common reason for technological difficulty during sessions related to the quality of the video connection and lag time between when the child or interventionist would speak/perform actions. This difficulty resulted in only one “dropped” video call session, which was completed via a phone call with the participant. The placement of the web camera was also a notable difficulty for participants. Four families had difficulty with ensuring that the webcam was positioned in a way that captured both the child and materials on the table during the first session. Out of these four families, only one family was not able to position the camera correctly after instruction and this resulted in the majority of sessions where the interventionist could not clearly see all of the child’s actions. The three other participants were fully visible after the first session. Other main technological concerns related to the audio connection while using the video conferencing software. Specifically, three participants had difficulty in connecting audio through their computer and web camera. For two of these families, the web camera was used to capture the video stream, however, audio for the sessions was conducted via a phone call. In total, 15% of all sessions across participants had some type of technological difficulty and 8% had to be rescheduled (see Table 2 for full details).

#### *Implementation and Completion of Sessions*

Eight of the ten children enrolled completed the intervention program. Two children who completed baseline assessments did not continue due to extenuating circumstances (time commitment did not work for one family and protocol was not appropriate for one child given current behavior challenges). Data will be reported for the eight children who completed the study protocol (baseline, intervention,

**Table 2** Program feasibility

<i>Technological considerations</i>	
Number of families able to use the video conferencing format	8/8
Number of sessions with technical difficulties	15/99
Number of session rescheduled due to technical difficulties	8/99
Most common technological difficulties families faced	Difficulty connecting webcam Difficulty setting up intervention space No audio connection Slow or lagged connection
<i>Implementation and completion of sessions</i>	
Mean length of intervention period	7.86 weeks
Mean length of session (#1–6)	16.67 min
Mean length of session (#7–12)	20.21 min
Most common barriers to successful session completion	Participant fatigue Too close to meal times Other distractors in the home (friends or family visiting)
Number of participants needing to reschedule sessions due to behavioral reasons	1/8

and post-intervention assessments). The mean number of sessions completed was 11.875 out of 12. One family did not complete all 12 sessions due to scheduling conflicts. Participation in the program ranged from 6 to 8 weeks, with an average length of 7.86 weeks. The increase in program length was due to rescheduling sessions because of last-minute schedule changes or planned family trips or vacations. Mean length of session was determined by calculating the time each participant was able to engage in the session with the interventionist (i.e. on-task behavior) minus the time the participant engaged in off task behaviors, such as engaging the interventionist in conversation or not playing through that session's story stem. Session start time was defined as when the interventionist began a story stem and the child focused their attention towards the interventionist. End time was defined as when the story stem came to a close and the child and interventionist cleaned up the toys. As can be seen in Table 2, for sessions 1–6, average time spent on task (or mean length of session) was calculated to be 16.67 min. The average length of session was also shown to increase during the intervention period across participants, with average on task time extending to 20.21 min for sessions 7–12, with peak session performance occurring during session #6–7.

### *Behavioral Considerations and Strategies*

The majority of participants needed some type of behavioral redirection or encouragement, given levels of fatigue or if the materials were specifically challenging during any given session. To help deter behavioral issues from the beginning of the intervention, during the first session, the interventionist let participants know that if, at any point, they chose not to participate, the session would be over, the interventionist would end the video call, and the child would not be able to see them for the full session time that day. This technique allowed the participant to know the “rules” of the intervention program (i.e., the child is required to engage in certain activities versus just talk to the interventionist), so if they did get upset, avoid the session content, or have a difficult time, they understood the consequences of their behaviors. Verbal reminders of this rule also helped participants re-engage into difficult sessions. Further, a review of the video recorded intervention sessions showed that the interventionists used positive praise, reward contingencies, redirection, and planned ignoring to help engage participants and keep them focused if any behavioral concerns (i.e., voicing fatigue or frustration, getting verbally upset) occurred. *Positive Praise* Every session included some form of positive praise. The interventionist would offer statements such as “great working,” “I like the way you are sitting in your seat and playing so nicely,” “You are providing such great ideas,” and “I know

this is hard work but you are doing great staying focused.” These statements helped all participants continue to engage in the intervention sessions. *Reward Contingencies* In some instances, a participant would have more difficulty during a session. During these sessions, which occurred across a few participants, the interventionist would introduce a reward that the child could work towards, such as reading a book or watching a TV show. For example, one participant wanted access to their favorite story book. The interventionist explained that if they were able to play first for 15 min then they could read their favorite book. A timer was also used in this situation to help the child know how much time they had left to play. If the child asked about how much time was left, they would be redirected to the timer. *Redirection* During some sessions, participants would also begin to talk about their school day or an upcoming event. In these instances, the interventionist would redirect the child to the content of the session and explain that if they completed the materials in the session, they could spend a few minutes afterward discussing their day or upcoming activities. *Planned Ignoring* Lastly, in a few instances, the participant would continue to talk or not fully engage in the session. During these moments, the interventionist would ignore their behavior and continue to play through the story or discuss the session content. This ignoring would typically lead the child to re-engage into the session, given that they were not gaining anyone's attention. As soon as the child began participating in the session, the interventionist would offer consistent positive praise.

During the intervention, if the participant seemed to have more difficulty engaging in a session, a post-hoc functional assessment was conducted on the recorded session to see if factors could be identified that would help explain that child's disengagement. Out of the eight participants, three required in-depth tape review. Post-hoc functional assessments showed that the largest participant factors impacting successful completion of sessions involved problem behaviors related to meal timing, fatigue, and motivation. As previous research has shown, children with PWS have difficulty with changes in routines or schedules, and given their dietary concerns, usually have set schedules surrounding meal times (Dykens et al. 1999; Dykens and Roof 2008). Given these concerns, every effort was made to avoid scheduling sessions within half an hour of an upcoming meal. Further, children with PWS partake in many services in addition to attending full school days, such as speech and language or physical therapy. Given these appointments, children can be quite fatigued by the end of a weekday. Care was also taken when scheduling to ensure that sessions occurred on days when the child had fewer activities. Even with these concerns in mind, two participants needed schedule changes during the intervention to ensure maximum participation. For both

participants, the interventionist worked with the parent in trying different days and times until the optimal session timing was obtained, when the child was neither distracted by approaching meal times, nor overtired from a busy day. Lastly, difficulty of the task and motivation to complete it also seemed to be a large factor in participation for three of the eight participants. Review of the recorded video sessions showed that these participants would voice “not wanting to play” and would engage in avoidance or escape behaviors (i.e., putting head down, engaging interventionist in conversation) when the interventionist pushed a difficult concept (i.e., name three emotions we could use in our story; what could we do to help this character feel less nervous?). In these instances, the interventionist took time to talk with the participant’s parents to conduct individual preference assessments as to what external motivators may help the child engage in the intervention sessions. In total, the three participants responded well to the implementation of a sticker or token system, where they would earn a sticker/token after engaging in a session. The accumulation of a certain number of tokens/stickers allowed the child to either access a preferred item (i.e., iPad time) or purchase a new one (i.e., puzzle set).

Overall, seven out of the eight participants did not have any sessions rescheduled due to behavioral concerns, and all eight participants were able to sit independently and engage with the interventionist via the video conferencing software without parental involvement. One participant needed to have two sessions rescheduled due to inability to engage in the session (i.e., putting head down, complaining of fatigue, and disconnecting from the video conferencing software) but was able to complete all 12 sessions in the program within 7 weeks.

Another factor that predicted participant engagement related to interest in the intervention toys. Given the aim to have a standardized intervention, all children were given the same set of toys that could not be changed or substituted during the play sessions. However, within the open feedback section of the BIRs, two parents voiced that the toys were not similar to the ones that their children typically engaged with. In these instances, the interventionists were able to re-engage the child to interact with the toys during these sessions. As described above, individual preference assessments on an as needed basis were completed to establish reinforcement systems for participants who had difficulty engaging in the sessions fully.

## Program Acceptability

### *Parent Perception of the PWS Telehealth Program*

Out of the eight participants to complete the program, seven caregivers completed the modified BIRs rating scale.

Based on survey results, caregivers gave an overall mean rating of 5.54/6.00 for the program’s acceptability. These items asked parents to report on if the content and structure of the sessions felt appropriate for the target population and in targeting goal skills. Specifically, all caregivers reported agreeing with the statement that “the intervention seemed appropriate for children ages 6–12 with PWS.” In terms of program effectiveness, caregivers gave a mean rating of 5.06/6.00 for the six items that reported on areas such as “the intervention may produce lasting improvements” or intervention effects on their child’s abilities in social cognitive domains. Importantly, all seven caregivers strongly agreed with the statement that “the intervention did *not* result in negative side-effects for my child.” Further, parent’s reported agreeing with the statement that “overall, the intervention was beneficial for my child.” Lastly, caregivers gave a mean rating of 4.86/6.00 for the program’s usability, which mostly reported on technological considerations, such as setting up the webcam, connecting to the online video conferencing software, and audiovisual considerations. Table 3 provides a full list of item results from the modified BIRs survey. Overall satisfaction was high (4.14/5.00) and parental attitudes towards telehealth as a modality through which treatment can be delivered was also rated positively (4.71/5.00). Several parents also noted that the sessions were an appropriate length in the open feedback section.

### *Parent Perception of Child’s Experience with the PWS Telehealth Program*

Parents who completed the BIRs also reported on their perceptions of how their child enjoyed engaging in the program. In terms of feasibility, it was not only important to establish if it was possible to engage the child over an online format, but also to understand if the child would enjoy and actively participate in this type of intervention structure. If children with PWS could not tolerate the intervention, even if parents’ viewed the program as feasible and beneficial, then it would have little practical implication moving forward. Results from the BIRs suggest that, overall, caregivers viewed their child as enjoying the intervention. Specifically, caregivers either agreed or strongly agreed with items relating to “I believe my child liked being part of the telehealth sessions” (5.71/6.00), “I believe my child liked interacting with the interventionist” (5.86/6.00), and “I believe my child would like to participate in a telehealth intervention again” (5.57/6.00). Parents who provided open-ended feedback specifically noted that their child looked forward to the sessions and enjoyed participating in them. Caregiver perception was also consistent with child behavior during sessions, as detailed above, in that the majority of participants completed all sessions



**Table 3** Treatment acceptability—modified BIRS

	Mean (SD)	Range
<i>Intervention acceptability &amp; effectiveness</i>		
Session structure seemed appropriate in targeting skills related to emotional, social, and imagination development	5.29/6.00 (0.49)	5.00–6.00
Session content seemed relevant to the goals of increasing play, social interactions, and flexible thinking	5.29/6.00 (0.49)	5.00–6.00
Intervention may produce lasting improvements in target skills	4.57/6.00 (0.98)	3.00–6.00
The intervention improved play, interactions, emotional understanding, or flexible thinking both during sessions and in other settings	4.29/6.00 (0.95)	3.00–6.00
The intervention did not result in negative side-effects for my child	6.00/6.00 (0.00)	6.00–6.00
The intervention seemed appropriate for children ages 6–12 with Prader–Willi syndrome	5.29/6.00 (0.76)	4.00–6.00
Overall, the intervention was beneficial for my child	5.14/6.00 (0.69)	4.00–6.00
<i>Child satisfaction</i>		
I believe my child liked being part of the telehealth sessions	5.71/6.00 (0.49)	5.00–6.00
I believe my child liked interacting with the interventionist	5.86/6.00 (0.38)	5.00–6.00
I believe my child would like to participate in a telehealth intervention again	5.57/6.00 (0.53)	5.00–6.00
I believe my child was able to see/hear the interventionist well	5.57/6.00 (0.53)	5.00–6.00
<i>Intervention usability</i>		
Setting up the webcam was straight forward and not time consuming	5.43/6.00 (0.79)	4.00–6.00
Connecting to the sessions was easy	4.57/6.00 (1.40)	3.00–6.00
I was frustrated with the video conferencing software	2.14/6.00 (1.68) <sup>a</sup>	1.00–5.00
Technology use did not seem to hinder our ability to have sessions	4.86/6.00 (1.35)	3.00–6.00
<i>Satisfaction with interventionist</i>		
I was satisfied with the interactions the interventionist provided my child	5.43/6.00 (0.53)	5.00–6.00
I believe the interventionist cared about my child	5.86/6.00 (0.38)	5.00–6.00
I believe the interventionist was knowledgeable	5.86/6.00 (0.38)	5.00–6.00
The interventionist provided good availability for sessions	6.00/6.00 (0.00)	6.00–6.00
<i>Overall satisfaction</i>		
I was satisfied with the interventionist's availability for communicating with me regarding the program, scheduling, and my child's behavior and progress	5.71/6.00 (0.49)	5.00–6.00
I found the parent coaching sessions useful	5.00/6.00 (0.82)	4.00–6.00
My overall attitude regarding intervention via telehealth is positive	4.71/5.00 (0.49)	4.00–5.00
My overall satisfaction with the telehealth program	4.14/5.00 (0.69)	3.00–5.00
<i>Comparison of telehealth to standard treatment methods</i>		
How do you believe telehealth compares with face-to-face interactions or therapies your child has been a part of?	3.33/4.00 (0.76)	2.00–4.00
What is the likelihood that you would recommend telehealth to other parents?	3.43/4.00 (0.53)	3.00–4.00

<sup>a</sup>Item is reversed scored (i.e., low scores indicate easier use of the video conferencing software)

without any difficulty and would voice “looking forward” to sessions and “being excited to play” with the interventionist even though the session content was difficult at times.

## Discussion

As research in rare disorders continues to progress, so too does a better understanding of the treatment needs of each population. However, with this increase in knowledge comes an equal increase in the difficulty of testing intervention procedures. Behavioral intervention work requires a large time commitment in the form of multiple in-person

sessions per week with a care provider. Given that this is not possible for most families who live far from clinic or hospital systems, alternative service delivery models need to be explored. Thus, the aim of this pilot study was to report on the feasibility and acceptability of a play-based telehealth intervention program for children ages 6–12 with PWS. Preliminary results suggest that telehealth is a feasible modality through which to deliver intervention directly to a child with PWS. Specifically, this study found that the majority of participants were able to engage in the intervention sessions independently, including playing with the interventionist across a computer screen, and complete the entire program with minimal behavioral or technological difficulty. Further, results showed that not only is it feasible

to deliver a play-based intervention directly to a child with PWS, but that the child also enjoyed the program and looked forward to participating in sessions. Lastly, results show that parents rated the program highly and believe it is a viable treatment option for children with PWS. While our pilot study does not address long-term benefits of intervention with this population, overall, parents reported that participating in the telehealth intervention did not cause any negative side effects, and was appropriate in targeting key skills relating to socioemotional development. These findings are extremely encouraging in terms of both using telehealth as a treatment option in PWS and in delivering intervention directly to a child with developmental delay through this modality.

The results from this study suggest that although overall parent report of their own and their child's experience in the program was high, some noteworthy considerations in terms of using telehealth specifically within the PWS population should be discussed. One of the primary considerations centers on troubleshooting technological issues. As was reported on the BIRs, caregivers felt as though connecting to the video conferencing software and maintaining a "glitch-free" connection was sometimes difficult and caused delays in either beginning the sessions or pauses within sessions. The tendency for the screen to freeze if there was a bad connection did cause some participants distress. Specifically, they would either call for their parents to help with the connection or become preoccupied with worrying if the screen would freeze instead of focusing on the session content. Usually, a simple redirection or explanation of what to do if the screen froze would allow the child to focus back on the interventionist. These situations also gave the interventionist an opportunity to work through feelings of distress with the child and help them use coping techniques discussed in the program. An important area for future work is continuing to better understand the use of telehealth in more rural areas, where internet connection may be very limited. A potential avenue to explore moving forward is the ability of families to complete sessions at a local library or school close to their home, which may have more reliable internet connection. This option does increase the burden of participating in sessions, but still allows for the family to access services at least within the comfort of their own community instead of having to travel long distances to partake in treatment opportunities.

Another issue to consider, particularly when working with the PWS population, is finding a conducive "space" and time for intervention sessions. Half of the families initially had difficulty angling the camera to capture both the child and the toys needed for each session. Further, it was important for families to find a place where the child would not be disturbed by either people or events going on (i.e., siblings wanting to be included, parents cooking

dinner) during the sessions. Results from this study suggest that an important first step, prior to beginning intervention sessions, may be to discuss with caregivers *where* the child will complete the sessions and *if* that space seems to be appropriate after sessions begin. This conversation may also include timing of the sessions, so as not to be too close to meal or snack times and when the child is not overly fatigued from his or her day. All of these factors greatly impact the child's baseline ability to engage in a session. Also, given that in using telehealth the interventionist does not have the ability to physically respond to the child as they would in-person, ensuring that these factors are addressed may naturally boost the efficacy of any program delivered via this modality, given that the child will be more likely to give full effort and attention.

Lastly, other main considerations include how to address any problem behaviors that the child evidenced, such as noncompliance or emotional upset, during intervention sessions. As research has shown, children with PWS display difficulties with emotion regulation and distress tolerance, along with rigidity in routines and perseveration (Cassidy et al. 2012; Dimitropoulos et al. 2013; Dykens and Roof 2008). During sessions, some of these behaviors evidenced themselves in terms of not wanting to play specific story stems (i.e., voicing disinterest in stories or toys; too fatigued to play) or complete the sessions. It is important to note that engaging in play, especially symbolic play, is more difficult for this population than their typically developing peers (Zyga et al. 2015). Asking a child with PWS to engage in more rigorous story stems or play more of an active role in building a storyline could have caused distress because it is a demanding task that may produce feelings of discomfort for the child. Thus, the child may have voiced feelings of disinterest in the toys or story stems as a way to try and avoid the difficult task. To address any noncompliance or verbal protests in the current study, the interventionists used behavioral techniques such as the use of timers, schedules, and positive reinforcement to engage the child in play for a certain time frame before then taking a break for a similar period of time. Over the course of the intervention, the time spent in play was continually increased for these participants and the break periods were subsequently decreased. As can be seen in Table 2, these techniques were effective in increasing the length of sessions and subsequent child participation from the first to the second half of the intervention program.

While these findings are quite promising, there are several limitations that should be noted. The small sample size may limit the generalizability of these preliminary findings to the PWS population. Further, results of this study do suggest that telehealth as an intervention option may be best suitable for those who are at least minimally verbal, able to attend to the interventionist without in-person

support, and do not have significant behavioral concerns. These limitations may not make telehealth a feasible option for the entire PWS population. However, given the rarity of PWS and the nature of this feasibility project, these findings are encouraging in that the majority of our participants were able to participate in the remote intervention. It should also be noted that parent report on intervention acceptability may be influenced by their knowledge of the aims of the current study and also the significant need for intervention in this community. The parents who participated in the current study knew that their children were engaging in the intervention to increase imaginative play, emotional understanding, and regulation abilities. This knowledge may have led to the Hawthorne effect and higher ratings on program satisfaction. Also important to note is that not all parents completed the BIRs survey and certain characteristics of the parents who did versus did not respond could impact overall reports of the intervention acceptability and feasibility. However, since it was an anonymous survey, the current paper cannot provide specific details on these parental characteristics. It is equally important to note though that the majority of parents did respond (7/8) and all parents provided some type of formal or informal feedback, which did not drastically differ (i.e. no one parent reported any significant dissatisfaction with the program). Further, given that PWS is a rare disorder, few behavioral interventions have been targeted specifically for PWS, and thus parents may be more likely to report positive outcomes in order to further develop intervention options in PWS. It is important to note, however, that parent report of acceptability was congruent with our other metrics for feasibility and acceptability reported here. Another important limitation to note is that while the BIRS has reported strong psychometric properties, the version used in the current study did have slight alterations in the form of adding further questions or word changes to ensure congruity with the aims of the current study. These changes alter the ability to fully rely on previously reported psychometric properties. However, other studies have modified the BIRS from its original structure (Wainer and Ingersoll 2013, 2015) and found similar overlap between behaviorally-based measures of feasibility and the given survey as is reported in this study. Lastly, an empirical question to consider, as it relates to next steps and potential limitations, is whether a first in-person visit is necessary or if the assessment and intervention could be delivered completely via remote technology.

Here we have established that long distance intervention via telehealth is feasible for children with PWS as young as 6 years of age. In particular, this study provides initial support that it is not only feasible for children with developmental delay or disorder to directly and independently participate in remote intervention sessions, but also that both the parent and child report satisfaction, and even

enjoyment, in engaging in this type of treatment format. Given these initial findings, important next steps are (1) to evaluate the efficacy of this intervention with respect to primary outcome variables on social-cognitive behavior and (2) to better understand the key “active ingredients” of the intervention program and for what age range and ability level this specific intervention is most suitable for. Long term aims include understanding how the use of telehealth can be expanded within the PWS population to other age ranges and ability levels. Taken together, these findings are extremely important not only for our study questions but also more broadly for future intervention work in PWS and other rare disorders. The ability to use telehealth successfully in rare disorders may greatly improve families’ connection to treatment resources earlier and more easily than before, placing these children on a more supported and upward course of skill growth and overall development.

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