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Systematic Review of Intervention Programs Designed to Improve the Socioemotional Skills of Children and Adolescents with Prader-Willi Syndrome

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Abstract:	The present study is a systematic review of intervention programs designed to improve the socioemotional skills of children and adolescents with Prader-Willi Syndrome (PWS). The search was conducted in the Web of Science and Pubmed databases following the PRISMA guidelines. A total of 6 studies made up the final sample and were organized based on the following psychological domains: social cognition and emotional competence. The findings suggest that these learning models may contribute to the development of socioemotional skills in children and adolescents with PWS. In addition, it appears that remote intervention through telehealth may be effective as a treatment option.

Systematic Review of Intervention Programs Designed to Improve the Socioemotional Skills of Children and Adolescents with Prader-Willi Syndrome

Abstract

The present study is a systematic review of intervention programs designed to improve the socioemotional skills of children and adolescents with Prader-Willi Syndrome (PWS). The search was conducted in the Web of Science and Pubmed databases following the PRISMA guidelines. A total of 6 studies made up the final sample and were organized based on the following psychological domains: social cognition and emotional competence. The findings suggest that these learning models may contribute to the development of socioemotional skills in children and adolescents with PWS. In addition, it appears that remote intervention through telehealth may be effective as a treatment option.

Key words: Prader-Willi Syndrome; intervention program; social cognition; emotional competence; theory of mind.

Introduction

The term Prader-Willi Syndrome (PWS) was first described by Langdon Down in 1887, and subsequently by Drs. Prader, Labhart and Willi in 1956 (Down, 1887; Prader et al., 1956). PWS is a rare or infrequent neurodevelopmental disorder that results from the loss of paternal expression of genes in the 15q11-q13 region (Tucci et al., 2019). About 60-70% of cases are caused by a deletion (DEL), 25-30% are due to maternal uniparental disomy (mUPD) (Butler et al., 2018) and approximately 4% of cases are caused by imprinting defects (Butler et al., 2019). PWS has an estimated incidence of 1:10000 to 1:30000 births and a prevalence of 1:10000 to 1:20000 persons in the United States (Bohonowych et al., 2019; Driscoll et al., 2017). While in Europe, the prevalence is estimated at 1:50000 inhabitants and the annual incidence at 1:30000 births (Gwenaëlle & Maithé, 2019).

Individuals with PWS are characterized by narrow foreheads, almond-shaped eyes, drooping corners of the mouth, and small feet and hands (Cassidy et al., 2012). Their development is marked by severe hypotonia and feeding deficits that begin in the neonatal period, followed by a period of hyperphagia and food obsession that often leads to severe obesity from childhood to adulthood (Poitou et al., 2023). They also have short stature due to growth hormone deficiency (Höybye & Tauber, 2022) and hypogonadism that can lead to incomplete pubertal development (Noordam et al., 2021).

From a cognitive perspective, individuals with PWS show mild to moderate intellectual disability (average IQ 60-70) comprising cognitive deficits, delayed motor and language development, and learning deficits (Höybye & Tauber, 2022). In addition, they often present difficulties in the mastery of executive functions, especially in planning, problem solving, working memory (Chevalère et al., 2015), attention and task switching (Woodcock et al., 2009). At the behavioral level, frequent symptoms include temper outbursts, impulsivity and rigidity, and obsessive-compulsive symptoms (Höybye & Tauber, 2022; Tunnicliffe et al., 2014).

In terms of social skills, individuals with PWS often present difficulties in maintaining friendships and getting along with others (Dimitropoulos et al., 2013; Dykens et al., 2017). These interpersonal difficulties are associated with them

presenting deficits in social cognition (Dykens et al., 2019), emotional competence (Famelart et al., 2022) and theory of mind (Lo et al., 2013).

Social cognition is a neuropsychological domain that is defined as the cognitive ability to adequately understand social situations and act accordingly (Henry et al., 2016). Individuals with PWS are at increased risk for difficulties with the following aspects of social cognition: reciprocal communication and interpretation of social cues (Dimitropoulos et al., 2013), facial and emotional discrimination (Dykens et al., 2019; Debladis et al., 2019), and imaginary (symbolic) play or pretend play (Dimitropoulos et al., 2019; Zyga & Dimitropoulos, 2020). Krasnor and Pepler (1980) defined pretend play based on four specific criteria: non-literalism, positive affect, intrinsic motivation and flexibility. Pretend play has been shown to relate to and impact areas such as social adaptation and communication, understanding others' emotional states, and socioemotional development (Barnett, 1990; Fehr & Russ, 2016; Singer & Singer, 1992).

Emotional competence refers to the ability to use emotions on a daily basis, including their expression, recognition, understanding and regulation (Mikolajczak, 2020). People with PWS present difficulties in recognizing and understanding basic emotions. In fact, they make on average 10% to 20% more errors in identifying and assigning emotions than the typical population, even when compared by developmental age (Whittington & Holland, 2017; Dykens et al., 2019). In addition, the emotional expressions of children with PWS are particularly poor and equivocal, making them difficult to interpret (Famelart et al., 2020).

Theory of mind is defined as a metacognitive and socioemotional process that enables an individual to understand one's own and others' emotions, beliefs and intentions (Premack & Woodruff, 1978). Children with PWS have less developed social skills, and often present difficulties in recognizing and understanding affective information and understanding others' point of view (Lo et al., 2013).

Findings from the literature argue that socioemotional competencies are useful as protective factors that facilitate the adaptation of the individual to the context and that favor better coping in the face of different life situations (Gutiérrez-Cobo et al., 2017; Mikulic et al., 2015). In the educational area, it has been evidenced that an adequate learning of socioemotional competencies favors the learning process, academic success

and problem solving (Bisquerra Alzina & Pérez Escoda, 2007; Gómez-Ortiz et al., 2017; Zins et al., 2004). In the area of mental health, it has been found that the development of socioemotional competencies contributes to the achievement of good psychosocial adjustment and emotional well-being favoring physical and mental health (Mayer et al., 2008; Pacheco & Fernández-Berrocal, 2013).

Systematic review is a methodological research tool that offers the possibility of updating and integrating the information available in the literature efficiently with systematic and explicit methods to identify, select and assess relevant research (Meca & Ausina, 2010; Perestelo-Pérez, 2013). Therefore, this tool could be very useful for the study of intervention programs aimed at promoting the socioemotional skills of the infant-juvenile population with PWS.

Considering that deficits in social cognition, emotional competence and theory of mind, together with behavioral problems are important determinants of the functioning and quality of life of the PWS population, an exhaustive systematic review of socioemotional skills intervention programs for children and adolescents with PWS was carried out. It is hoped that the findings will provide learning models by which children or adolescents with PWS can improve their social cognition, emotional competence, and socioemotional skills, with which to acquire greater psychosocial adjustment and quality of life. Likewise, it is hoped that the systematic review will provide an understanding of the current state of socioemotional skills intervention programs, so that future lines of research can design and develop intervention programs in accordance with the needs of the infant and adolescent population with PWS.

Research questions

To provide a comprehensive overview of existing intervention programs and their outcomes, the analysis in this review aimed to answer the general review questions "what works, for whom and under what circumstances" (Popay et al., 2006).

The first research question addressed the effects of the intervention program on the target population "what works for whom".

1. What is the existing evidence on intervention programs that are effective in improving the social cognition, emotional competence and theory of mind of children and adolescents with PWS?

The second research question provided an overview of existing intervention programs and contextual factors to clarify "what circumstances" fostered intervention effects.

2. How do contextual factors, such as the type of intervention (face-to-face or online) and the sample (size and inclusion/exclusion of control group) affect the effect of the intervention?

Method

Design

A systematic review was conducted following the PICOS criteria (Table 1) and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Page et al., 2021). The study protocol has been registered in the Open Science Framework (OSF) (Associated project: osf.io/2znaf).

Table 1

Search strategy according to PICOS criteria (Population; Intervention; Comparison; Outcome; Study design)

P	Children and adolescents diagnosed with PWS.
I	Intervention programs that assess socioemotional skills.
C	Any group comparisons using normative data for comparison.
O	Performance of socioemotional skills (longitudinal studies).
S	Empirical intervention programs (any sample size).

Eligibility Criteria

The inclusion and exclusion criteria for the present systematic review are presented in Table 2.

Table 2

Inclusion and exclusion criteria for the systematic review

Inclusion Criteria	Exclusion Criteria
- Participant: children or adolescents with PWS. - Type of study: empirical and longitudinal intervention programs.	- Accessibility: full text not available. - Type of study: - Cross-sectional studies. - Single-case studies. - Review studies.

- Assessment: psychological or neuropsychological tasks or tests to assess socioemotional skills. - Publication date: studies published between 2014 and 2024. - Language: studies published in English or Spanish.	- Training intervention programs for carers of children and adolescents with PWS. - Intervention programs aimed at the adult population with PWS. - Thematic: no reference to socioemotional aspects.

Search strategies

A literature search was conducted to identify intervention programs that assessed the socioemotional skills of children and adolescents with PWS. The search was conducted in December 2024 in the Web of Science and PubMed databases. The search terms used were (a) Prader-Willi syndrome, (b) socioemotional skills, (c) social skills, (d) social cognition, (e) theory of mind, (f) emotional competencies, (g) intervention, (h) strategies, (i) program, (j) therapy, and (k) proposal. Table 3 shows the combination of these keywords. A filter was applied with respect to the year of publication (2014-2024) in both databases.

Table 3

Search strategies

PudMed
(Prader Willi Syndrome[Title/Abstract]) AND (socioemotional skills[Title/Abstract]) AND ((intervention[Title/Abstract]) OR (strategies[Title/Abstract]) OR (program[Title/Abstract]) OR (therapy[Title/Abstract]) OR (proposal[Title/Abstract]))
(Prader Willi Syndrome[Title/Abstract]) AND (social skills[Title/Abstract]) AND ((intervention[Title/Abstract]) OR (strategies[Title/Abstract]) OR (program[Title/Abstract]) OR (therapy[Title/Abstract]) OR (proposal[Title/Abstract]))
(Prader Willi Syndrome[Title/Abstract]) AND (social cognition[Title/Abstract]) AND ((intervention[Title/Abstract]) OR (strategies[Title/Abstract]) OR (program[Title/Abstract]) OR (therapy[Title/Abstract]) OR (proposal[Title/Abstract]))
(Prader Willi Syndrome[Title/Abstract]) AND (theory of mind[Title/Abstract]) AND ((intervention[Title/Abstract]) OR (strategies[Title/Abstract]) OR (program[Title/Abstract]) OR (therapy[Title/Abstract]) OR (proposal[Title/Abstract]))
(Prader Willi Syndrome[Title/Abstract]) AND (emotional competences[Title/Abstract]) AND ((intervention[Title/Abstract]) OR (strategies[Title/Abstract]) OR (program[Title/Abstract]) OR (therapy[Title/Abstract]) OR (proposal[Title/Abstract]))
Web of Science

(TI=(Prader-Willi Syndrome)) AND (TS=(socioemotional skills)) AND (TS=(intervention) OR TS=(proposal) OR TS=(program) OR TS=(strategies) OR TS=(therapy))
(TI=(Prader-Willi Syndrome)) AND (TS=(social skills)) AND (TS=(intervention) OR TS=(proposal) OR TS=(program) OR TS=(strategies) OR TS=(therapy))
(TI=(Prader-Willi Syndrome)) AND (TS=(social cognition)) AND (TS=(intervention) OR TS=(proposal) OR TS=(program) OR TS=(strategies) OR TS=(therapy))
(TI=(Prader-Willi Syndrome)) AND (TS=(theory of mind)) AND (TS=(intervention) OR TS=(proposal) OR TS=(program) OR TS=(strategies) OR TS=(therapy))
(TI=(Prader-Willi Syndrome)) AND (TS=(emotional competencies)) AND (TS=(intervention) OR TS=(proposal) OR TS=(program) OR TS=(strategies) OR TS=(therapy))

Note. TI = title; TS = topic.

Selection of studies

A preliminary search identified a total of 41 studies. This first study selection process was carried out by the first author of the study (A.P.). After eliminating 4 duplicate studies, 37 were reviewed by two independent authors (P.P. and S.A.) who analyzed titles and abstracts. When there was a disagreement, a fourth reviewer helped to achieve a consensus (J.F.L.P.). After screening and reviewing for inclusion and exclusion criteria, a total of 6 studies were selected for full review (A.P., P.P., S.A. and J.F.L.P.). In total, 6 papers were included in this systematic review. The detailed selection process is illustrated in Figure 1.

Data extraction

Data were collected on the 6 studies included in the systematic review. The following variables were analyzed: author and name of the program, sample size, intervention program, psychological tests or tasks, and results. This data collection process was carried out by the first author of the study (A.P.). After that, two independent authors (J.F.L.P. and O.M.) reviewed and confirmed the data.

Risk of bias

The modified Newcastle-Ottawa Scale (Bawor et al., 2014) was used to evaluate the risk of bias of the included studies. The aim of this scale is to examine the quality of each study in the following domains: sample selection (selection bias), control confounding (conduct bias), statistical method (detection bias) and outcome measures (reporting bias).

Results

Initially, 41 studies were found, of which only 6 studies were selected that met the eligibility criteria.

Analysis of study quality and potential biases in the eligible articles revealed shortcomings in sample-related methods. To strengthen future research, these methods should be enhanced. Key challenges for improvement include the need for larger participant groups (Table 4).

Table 4

Risk of bias assessment for reviewed studies

	Method for selecting sample	Methods to control confounding		Statistical methods		Methods for measuring outcomes	
		Sample size	Identification of confounders	Appropriate analyses	Missing data	Outcome measures	Objective assessment
Dykens et al., 2022	Moderate	Moderate	Moderate	Low	Low	Low	Low
Vascelli et al., 2023	Moderate	High	High	Moderate	Moderate	Low	Moderate
Dimitropoulos et al., 2021	Moderate	High	Moderate	Moderate	Low	Moderate	Low
Dimitropoulos et al., 2022	Moderate	High	Moderate	Low	Low	Low	Moderate
Dimitropoulos et al., 2024	Moderate	High	Moderate	Low	Low	Low	Moderate
Famelart et al., 2022	Moderate	High	Moderate	Moderate	Low	Moderate	Moderate

Note. The modified Newcastle-Ottawa scale (Bawor et al., 2014) has been used to analyze the risk of bias of the studies reviewed. High risk of bias: High; Moderate risk of bias: Moderate; Low risk of bias: Low.

Table 5 shows the studies on intervention programs designed to develop socioemotional skills in children and adolescents with PWS. The table shows different characteristics of the studies, such as the author and name of the program, the sample size, the description of the intervention analyzed, the psychological tests or tasks used and the significant results found.

The results of the systematic review showed the efficacy of 6 intervention programs designed to improve the socioemotional skills of children and adolescents with PWS. From the review of these studies, the results were organized based on the following psychological domains: social cognition (N=5), composed of studies on socialization and communication skills (N=2) and pretend play skills (N=3); and emotional competence (N=1). The results for each domain are presented below.

Social cognition

Socialization and communication skills

We found two online intervention programs that aimed to improve social and communication skills of adolescents with PWS.

Dykens et al. (2022) developed the BOSS (Building Our Social Skills) online social skills program in which 51 adolescents and young adults with PWS participated. The 30-minute sessions were group-based (6-8 participants) and ran 3 times per week for 10 weeks using Zoom. The results showed an interesting effect of the program on socialization. The participants acquired social skills, as well as a greater number of friends and contact with them, thus reducing their feelings of loneliness. However, the study did not include a control group, which limits the effectiveness of the intervention.

Vascelli et al. (2023) designed an online socialization program in which 2 adolescents with PWS conversed with elderly and adolescents selected from the Ciciarapp program (Berardo et al., 2020; Berardo et al., 2021), with whom they were to have conversations. Conversation sessions with an average duration of 30 minutes were held once a week via Skype. Two specific training phases were implemented. In the first phase, a 1-second delay was used to present the textual message, whereas, in the second phase, a 5-second delay was included. The number of initiations and follow-up questions (responses) were taken into account. The results of the study suggested that participants improved their social communication skills during online socialization with their partners. In addition, the number of spontaneous responses increased significantly in the second phase. However, the study had a very small sample size and also did not include a control group.

Pretend play skills

We also identified three studies assessing the efficacy of three telehealth intervention programs based on symbolic play aimed at developing socioemotional skills in children with PWS.

Dimitropoulos et al. (2022) conducted a follow-up of the initial PRETEND program feasibility and acceptability of telehealth in a training intervention for parents of children with PWS (Zyga et al., 2018). They reported the preliminary efficacy of the program in improving pretend play skills and cognitive and social skills in preschool

children with PWS through two studies. Study 1 described the pilot parent training program in a format of 12 sessions of 30-45 minutes 2 times per week in remote. Study 2 refined the program by directly involving the children. There were 8 sessions of 45-60 minutes delivered 1 time per week, and included 3 live play sessions of 15-20 minutes and play tasks assigned to the parent-child dyads. The PRETEND program demonstrated efficacy when play sessions with live coaching incorporated children into the intervention. Specifically, increased time spent in play and significant improvements in cognitive and affective play skills were observed in children with the mUPD subtype who underwent the intervention compared to children with wait-listed mUPD. However, the study sample lacked diversity and was quite small, making it difficult to generalize the results to a larger population of preschool children with PWS.

Dimitropoulos et al. (2021) conducted a follow-up study on feasibility and acceptability of telehealth for direct intervention in children with PWS (Dimitropoulos et al., 2017), and reported on the effectiveness of this program based on symbolic play. The program included 15-20 minutes sessions, 2 times per week for 6 weeks in which the interventionist played with the child via videoconferencing. The program worked on play skills, emotional expression and recognition, and behavioral self-management techniques. In addition, parents had 3 additional 5-10 minute sessions with their children. Overall, the program was effective in improving the children's social, emotional, cognitive and behavioral functioning. The children obtained significantly improved cognitive and affective processes in pretend play, and an increase in their cognitive flexibility and divergent thinking fluency. However, the lack of a control group limits the implications that can be made about the efficacy of the intervention.

Dimitropoulos et al. (2024) conducted a new intervention program in which they added the waiting list control group condition to further support the efficacy of the telehealth intervention program based on symbolic play (Dimitropoulos et al., 2021). The program included 8 weekly direct play sessions between the interventionist and the chil via videoconference, and 4 biweekly parent education sessions and parent-child joint play sessions. Participants who received the intervention demonstrated significant improvements in the organization of pretend play and in the frequency of affect expression, positive social interactions, and number of themes exhibited during play. However, the sample size was relatively small and treatment-related differences among PWS genetic subtypes were not assessed, making it difficult to generalize the results.

Emotional competence

Famelart et al. (2022) designed an intervention program to directly improve emotional skills in children with PWS. The study showed that children who participated in the EMO-T program of EC training for 6 weeks showed significant and sustainable improvement in the ability of voluntary expression, recognition and understanding of emotions, matching the baseline level of children with typical development (TD). Likewise, they improved in emotion regulation, despite not receiving specific training in that skill. However, the study had a small sample size and did not include a standardized methodology for direct observations.

Discussion

The results of this systematic review indicate the efficacy of 6 intervention programs designed to improve the social and affective skills of children and adolescents with PWS.

The EMO-T program (Famelart et al., 2022) was effective in improving all aspects of emotional competence, including emotional regulation skills despite not being directly exercised in the program. These results support the developmental model of emotional competence, which shows that emotion regulation skills require prior emotional skills (expression, recognition and comprehension skills) and should be more fully considered in future intervention programs (Famelart et al., 2020; Famelart et al., 2022).

Two studies reported the efficacy of two online intervention programs aimed at developing socialization and communication skills. The BOSS intervention was practical and well tolerated in improving social skills, promoting friendships, and reducing feelings of loneliness in adolescents with PWS (Dykens et al., 2022). The online socialization program with elderly and adolescent conversation partners (Vascelli et al., 2023) indicated improvements in conversational skills and an increase in the number of spontaneous responses in adolescents with PWS during the second intervention phase. Through these programs, adolescents with PWS can acquire social skills that allow them to improve their interactions and increase their friendships. However, future lines of research should focus on dessigning interventions that reduce

feelings of loneliness due to its negative consequences for mental health and well-being, especially in individuals with intellectual disabilities.

Three studies indicated promising findings on three symbolic play-based telehealth intervention programs aimed at developing socioemotional skills in children with PWS. Dimitropoulos et al. (2021) found that children acquired significantly improved cognitive and affective processes in the pretend play, and an increase in their cognitive flexibility and divergent thinking fluency after the intervention. Dimitropoulos et al. (2024) observed that several cognitive and affective processes in pretend play improved after intervention, such as organization in play, frequency of affect expression, positive social interactions, and the number of themes exhibited in play. Similarly, the PRETEND program was effective in developing cognitive and affective play skills (Dimitropoulos et al., 2022). These interventions are effective not only in improving children's individual pretend play skills, but also in improving children's emotional and behavioral understanding and functioning through the use of play.

In relation to symbolic play, evidence suggests that play behavior may vary as a function of genetic subtype in individuals with PWS. Differences in cognitive, social, and behavioral skills between the two main PWS subtypes and parental characteristics, such as stress, may result in responding differently (Hartley et al., 2005; Hogart et al., 2010; Ihara et al., 2014; Whittington et al., 2010). Dimitropoulos et al. (2019) found that children with the DEL subtype scored higher than children with the mUPD subtype and Autism Spectrum Disorder (ASD) in the organization and time spent in symbolic play and in the frequency with which they incorporated affect into play. However, Dimitropoulos et al. (2022) using the PRETEND program among different genetic subtypes of PWS found increased time spent in play and significant improvements in cognitive and affective play skills in children with the mUPD subtype who underwent the intervention, but not in children with the DEL subtype. Thus, it appears that future interventions for this population should be tailored to each genetic subtype in order to obtain similar improvements in functioning.

It is noteworthy that five programs employed a remote or online methodology in their interventions rather than traditional face-to-face treatment. So, it appears that remote intervention via telehealth may serve as a promising alternative to face-to-face

treatment in the PWS population. In fact, it is becoming increasingly prevalent in the rare disease community (Cox et al., 2012; McGaery et al., 2012). Dimitropoulos et al. (2017) in a pilot study reporting on the feasibility of using telehealth for direct intervention to 8 children aged 6-12 children with PWS found promising results in terms of both the use of telehealth as a treatment option in PWS, as well as direct intervention to a child with developmental delay. Similarly, Zyga et al. (2018) supported the use of telehealth in a training intervention to parents of children with PWS aged 3 to 6 years.

All three symbolic play-based intervention programs support the use of telehealth. Overall, studies evidence that remote intervention delivered directly to the child can be effective in increasing pretend play skills in children with PWS (Dimitropoulos et al., 2021; Dimitropoulos et al., 2022; Dimitropoulos et al., 2024). Given that play skills are associated with socioemotional and sociocognitive functioning, the findings of these studies indicate an important new avenue of feasible and accessible behavioral intervention for school-aged children with PWS.

In terms of limitations, it should be noted that the number of studies selected in the systematic review was very small. The search strategy was conducted in the Web of Science and PubMed databases, which were considered to guarantee the principles of completeness and relevance, respectively. However, it is possible that a greater number of intervention programs could have been obtained with other databases or by relaxing the inclusion and exclusion criteria. Nevertheless, since PWS is a rare disease, very few interventions have been performed to date in this population. Therefore, it is necessary to take into consideration these studies that showed benefitial results by which children and adolescents with PWS can acquire a learning model to improve their socioemotional skills and achieve greater psychosocial adjustment.

All the studies evidenced beneficial results in the social cognition and emotional competence of children and adolescents with PWS. However, none of them studied theory of mind. Therefore, it would be desirable that future intervention programs address this construct so that individuals with PWS can acquire a better understanding of affective information and of their own and others' beliefs and intentions.

It should be mentioned that one of the studies included in the systematic review, the BOSS program, was not only aimed at adolescents but also at young adults

(M=20.8) (Dykens et al., 2022). However, it was decided to include it in the systematic review because it was aimed at adolescents and because the program addressed the constructs that were the focus of the study, coinciding with the inclusion criteria.

Another limitation to consider is that three of the articles of the systematic review did not include a control group. However, rare population trials generally have smaller sample sizes and tend to use single-group, non-randomized, unblinded designs compared to trials in common diseases (Bell & Tudur Smith, 2014). The remaining three articles employed a control group that received the intervention at the end of the protocol. Nevertheless, it is worth mentioning that using a placebo control group could help to obtain a more accurate and rigorous evaluation of the intervention, as it is considered a possible and effective treatment in psychological interventions (Gaab et al., 2019; Zhu et al., 2014).

Therefore, more research is needed in this field that includes interventions with larger sample sizes and greater scope to increase the efficacy and generalizability of the results found. Likewise, it is important that future studies incorporate a randomized control group, if possible that received a placebo intervention, to confirm that any observed outcome is the direct result of the intervention. In addition, without a control group, the possible influence of developmental change over time is unknown. Future studies should also take into account the genetic subtype of PWS when carrying out the intervention. On the other hand, very little is known about the long-term maintenance of improvements in socioemotional skills after these interventions, so it would be advisable to conduct post-intervention sessions over time. Future lines of research should take these considerations into account to improve the quality and validity of the findings.

Conclusion

The results of the systematic review demonstrate the efficacy of 6 intervention programs as a viable treatment option for the development of socioemotional skills in children and adolescents with PWS. The EMO-T program was effective in developing and improving emotional competence (Famelart et al., 2022), while the conversation program with elders and adolescents (Vascelli et al., 2023) and the BOSS intervention (Dykens et al., 2022) were beneficial in improving socialization and communication

skills in adolescents with PWS. Pretend play-based interventions proved a promising avenue for acquiring improvements in social, emotional, cognitive, and behavioral functioning (Dimitropoulos et al., 2021; Dimitropoulos et al., 2022; Dimitropoulos et al., 2024). Furthermore, it appears that remote intervention through telehealth may be effective in implementing socioemotional skills.

These findings suggest that these learning models may contribute to the development of social cognition and emotional competence in children and adolescents with PWS. However, further research, including larger sample sizes, control groups, and long-term follow-up, is needed to determine whether these gains translate into lasting improvements in psychosocial adjustment and quality of life.

Declarations

Conflict of interest The authors declared that they had no conflict of interest.

Data availability No data was used for the research described in the study.

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Table 5

Results of the review of studies on intervention programs designed to develop socioemotional skills in children and adolescents with PWS

Authors	Program	Sample	Intervention	Psychological test/task	Results
Dykens et al., 2022	BOSS online social skills program.	51 adolescents and young adults with PWS (M=20.8) and their parents.	Group sessions (6-8 participants) of 30 minutes, 3 times a week, for 10 weeks using Zoom, and 3 month follow-up.	Social Responsiveness Scale-2 (SRS-2) Child Behavior Checklist (CBCL)	Adolescents improved their social skills, got along better with their peers, improved the quality of their interactions, had more friends and less loneliness.
Vascelli et al., 2023	Online socialization program with elderly and adolescent conversation partners.	2 adolescents with PWS (19 years), and 4 elderly and 2 adolescents selected from the Ciciarapp Program.	30 minute conversation sessions, once a week via Skype. Phase 1: 1-second delay to present the textual message. Phase 2: 5-second delay.	Vineland Adaptive Behavior Scales (VABS) Reinforcement Inventories for Children and Adults (RICA) UCLA Loneliness Scale School Intervention Rating Form (SIRF)	Adolescents improved their social communication skills. The number of spontaneous responses increased significantly in Phase 2 in adolescents with PWS.
Dimitropoulos et al., 2021	Telehealth intervention program based on the pretend play.	15 children with PWS (6-12 years) and their parents.	15 minute sessions, twice a week for 6 weeks between interventionist and child via videoconference. 3 additional sessions of 5-10 minutes between parent and child. Post-intervention session after 4 weeks.	Kaufman Brief Intelligence Test – Second Edition (KBIT-2) Vineland Adaptive Behavior Scales, Second Edition (Vineland – II) Affect in Play Scale (APS) Multidimensional Stimulus Fluency Measure (MSFM)	Children demonstrated significantly improved cognitive and affective processes in the pretend play. Cognitive flexibility and divergent thinking fluency increased in children with PWS.
Dimitropoulos et al., 2022	PRETEND program based on the remote symbolic play.	Study 1: 30 children with PWS (3-5 years) divided into: intervention group (n=15) and wait-list control group (n=15), and their parents. Study 2: 18 children with PWS (3-5 years) divided into: intervention mUPD (n=6), wait-list mUPD (n=6), intervention DEL (n=4) and wait-list DEL (n=2), and their parents.	Study 1: 12 parent training sessions of 30-45 minutes, 2 times a week remotely. The child was not directly involved. Post intervention session at 4 weeks. Study 2: 8 sessions of 45-60 minutes once a week. 3 live play sessions of 15-20 minutes and play tasks between parent and child. Post intervention session after 4 weeks. At the end of the protocol, the waiting list control groups received the intervention.	Mullen Scales of Early Learning (MSEL) Peabody Picture Vocabulary Test (PPVT-4) Social Skills Improvement System Rating Scales (SSIS) Social Communication Questionnaire (SCQ) Parenting Stress Inventory (PSI-4) Affect in Play Scale – Preschool (APS-P)	The PRETEND program demonstrated efficacy when live-training play sessions incorporated children into the intervention. Significant improvements in cognitive and affective play skills, and increased time spent playing, in children with the mUPD subtype who underwent the intervention compared to those on the waiting list.
Dimitropoulos et al., 2024	Remote intervention based on the pretend play.	19 children with PWS (6-9 years old) divided into: intervention group (n=10) and waiting list control group (n=9), and their parents.	8 weekly sessions of direct play between interventionist and child via videoconference. 4 biweekly parent education sessions. 4 biweekly sessions of joint play between parent and child. At the end of the protocol, the waiting list control group received the intervention.	Vineland Adaptive Behavior Scales, Second Edition (Vineland–II) Social Skills Improvement System Rating Scales (SSIS) Parenting Stress Inventory (PSI-4) Social Communication Questionnaire (SCQ) Affect in Play Scale (APS) Kaufman Brief Intelligence Test – Second Edition (KBIT-2)	Participants who received the intervention demonstrated significant improvements in the organization of pretend play and in the frequency of affect expression, positive social interactions, and number of themes exhibited in the game.

Famelart et al., 2022	EMO-T emotional competence training program.	25 children with PWS (5-10 years) divided into: experimental group (n=13) and control group (n=12), and 50 Typically Developing (TD) children (3-10 years) matched with PWS by sex and chronological age (n=25) and by sex and age of intellectual development (n=25).	1 pre-intervention assessment session for all children. Weekly 30 minute sessions for 6 weeks to children with PWS in the experimental group and their regular therapist. Children with PWS in the control group continued with their usual rehabilitation program. 2 post-intervention sessions to all children with PWS, at the end of the program and 3 months later. At the end of the protocol, children with PWS in the control group received the intervention.	EMOrea Task EMOmim Task Identification Task Matching Task Naming Task Affective Judgment Questionnaire (AJQ) Emotion Regulation Checklist (ERC-French version)	PWS children in the experimental group showed significant and sustainable improvement in emotion expression, recognition and comprehension skills, matching the baseline level of all TD children. PWS children in the experimental group improved in emotion regulation, although they did not receive specific training in that skill.

Figure 1. A PRISMA flow diagram of the search strategy for studies on socioemotional skills intervention programs for children and adolescents with PWS.