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ORIGINAL PAPER

Behavioral and Dietary Management for Adults with Prader–Willi Syndrome in a Residential Setting

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Abstract

Objectives We present a program evaluation wherein archival data collected at The Arc of Alachua County (The AArc) was evaluated to determine the effects of a comprehensive treatment package on the management of weight and behavior in adults with Prader-Willi syndrome (PWS).

Methods Data from 4 individuals with PWS who participated in The AArc's Program over a span of 6 years were collected. Data were analyzed in four phases: a descriptive analysis providing a summary of the characteristics of the individuals, a two-tailed paired sample *T* test to evaluate the change in weight, analyses of behavioral topographies using Wilcoxon's matched pairs tests, and a calculation of clinical significance using percent-reduction for behavioral topographies inappropriate for statistical analysis due to small sample sizes.

Results Analyses showed a statistically significant reduction in weight, food stealing, tantrum behavior, skin picking, and self-injury. Behavior not included in the statistical analysis due to small sample sizes showed clinically significant reductions.

Conclusions Although the analyses were conducted with archival data, the treatment package implemented in The AArc's PWS program suggests strong historical evidence to support efficacy in reducing weight, food stealing, and other behavioral topographies. Recommendations for future research are discussed.

Keywords Prader–Willi syndrome · Behavior management · Tantrum · Weight management · Food stealing

Prader–Willi syndrome (PWS) is a complex neurodevelopmental syndrome that affects between 1 in 15,000 and 30,000 people worldwide (Cassidy et al. 2012). PWS is characterized by mild to moderate intellectual impairment, hypotonia (low muscle tone), hypogonadism (malfunction of the gonads), delays in language and motor development, short stature, and hyperphagia (excessive hunger) (Cassidy et al. 2012). The presence of hyperphagia often leads to aggressive food-seeking, food-stealing, and over-eating that can result in immediate death due to stomach rupture. Hyperphagia can also predispose individuals with PWS to develop obesity and associated health risks, including cardiovascular issues, respiratory issues including sleep apnea, and diabetes (Cassidy et al. 2012; Dykens 2000).

In addition, individuals are likely to present with behavioral and psychiatric disturbances, which can begin as early as 2 to 6 years of age and persist throughout adulthood (Cassidy and Driscoll 2009; Dykens 2004). Common behavioral problems include food stealing, skin picking, compulsive behaviors including hoarding, difficulty with changes in routine, and tantrum behaviors (Cassidy and Driscoll 2009; Symons et al. 1999). Approximately 15% of individuals also engage in rectal digging (Hanchett 1994). While rectal digging was previously thought to be maintained solely by attention and automatic reinforcement in the form of self-stimulation, anecdotal evidence from practitioners in recent years suggests automatic negative reinforcement related to escape from constipation as a possible function (L. Ramos, personal communication, May 15, 2018).

The occurrence of challenging behavior combined with hyperphagia often means that individuals with PWS require close monitoring and intensive intervention to prevent life-threatening consequences (Cassidy et al. 2012). There have

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been extensive attempts to manage symptoms using medical/surgical, pharmacological, and behavioral interventions, each with varying degrees of success. Collective data on gastric bypass, laparoscopic sleeve gastrectomy, and biliopancreatic diversion demonstrate that hyperphagia in individuals with PWS cannot be remedied with surgical manipulation of the gastrointestinal tract alone. These surgical interventions are not recommended due to failure to curb appetite, dangerous side effects, and extensive weight gain observed during long-term follow ups (Arnold 2017; Liu et al. 2020; Marinari et al. 2001; Scheimann et al. 2012).

To date, no medication has demonstrated a clinically significant long-term effect on eating behaviors or obesity in PWS, and the results of studies examining the effects of medication aimed at treating behavior problems show widely varying and often conflicting results (Bonnot et al. 2016; Driscoll et al. 2020; Miller et al. 2015). Human growth hormone has proven integral in increasing muscle mass, height, and intellectual ability, especially when treatment is started early; however, it does not reduce hyperphagia, prevent obesity, or reduce behavior problems (Butler et al. 2013; Höybye 2013; Lo et al. 2015). Oxytocin trials produced inconsistent results on whether or not the medication is effective for treating hyperphagia, food-related behaviors, and social behaviors (Kuppens et al. 2016; Miller et al. 2017). Some research even indicates an increase in tantrum behaviors on higher doses of oxytocin (Einfeld et al. 2014).

Psychotropic medications have been extensively researched as a method for controlling both hyperphagia and behavior problems (Bonnot et al. 2016; Driscoll et al. 2020). Atypical antipsychotics, including risperidone have been suggested as a treatment for behavior problems and appetite in PWS. Although this class of medication appears effective for psychotic symptoms, results implicating uses for hyperphagia and behavior problems are preliminary and require replication (Bonnot et al. 2016). Selective serotonin reuptake inhibitors have demonstrated some effect on managing mental health symptoms, including obsessive-compulsive behaviors; however, additional research is needed to report conclusively (Bonnot et al. 2016; Driscoll et al. 2020). Research on topiramate is similarly inconclusive. There is conflict regarding whether or not the medication has an effect on hyperphagia, skin picking, tantrum behaviors, or obsessive–compulsive disorder (Bonnot et al. 2016; Consoli et al. 2019; Shapira et al. 2002).

Behavioral interventions have shown some promising effects on weight, food stealing, and behavior problems. Reinforcement for exercise, reduced calorie consumption, and weight loss have all been used successfully within treatment packages for weight reduction (Altman et al. 1978; Page et al. 1983; Singh et al. 2008, 2011). Teaching discrimination between healthy and unhealthy food or safe and forbidden foods, and education on informed meal choices have shown some effect on reducing covert food stealing and weight (Maglieri et al. 2000; Singh et al. 2008, 2011). Mindfulness has been included in several very successful treatment packages for managing both weight and aggression (Singh et al. 2008, 2011, 2017). Differential reinforcement of other behaviors (DRO), a procedure where reinforcement is delivered on a specific schedule contingent on the absence of a specific behavior, has proven effective in reducing covert food stealing and weight in individuals with PWS (Cooper et al. 2020; Page et al. 1983, b; Rone 2010). Additionally, a treatment package including DRO, limited bathroom time, and functional communication training (FCT), a procedure where a functionally equivalent communicative response is taught to replace a problem behavior, demonstrated some success in eliminating severe rectal digging in a 26-year-old man with PWS (Stokes and Luiselli 2009).

The existing literature on managing weight and behavior within PWS is severely limited, especially in terms of behavior analytic interventions. There is currently no definitive best practice for comprehensive symptom management; however, a combination of pharmacological and behavioral treatments may prove beneficial. The purpose of this paper is to describe a medical–behavioral program for the management of symptoms associated with PWS, which includes medical, pharmacological, and behavioral interventions, and to use archival data to evaluate the long-term effects of the program on weight and problem behaviors. Although the information gained from archival data is limited, particularly with regard to the causal effects that can be drawn about each component of the treatment package, this article is aimed at describing a treatment package that shows promising long-term results in the hopes of prompting additional research to identify the effects of each component in order to improve future treatment.

Methods

Participants

Data from 24 women and 21 men with PWS between the ages of 20 and 57 were selected for inclusion in this study. All participants resided at The AArc, a residential facility that offers a comprehensive program for the management of weight and behavior problems associated with PWS. The selection process required individuals to be at least 18 years of age and have a diagnosis of PWS. Additionally, participants were required to have resided in The AArc's PWS program for at least six continuous years at the time of data collection. Given these parameters, 13 individuals were excluded as they had resided in the program for fewer than 6 years or had been absent from the program for over six consecutive months. The result was the availability of full data sets for 45 individuals.

Procedures

Below is a description of the treatment package implemented at The AArc during the 6-year period of data collection. The homes were specially equipped to include relevant antecedent conditions to prevent food stealing, such as locked or alarmed kitchens and pantries. Specialized treatment staff members provided 24-h supervision, most often at a ratio of at least one staff member for every three individuals, with rare exceptions. The program was highly structured to provide appropriate nutrition and meal distribution to promote the individuals' natural metabolisms and reduce the likelihood of common adverse health concerns such as osteoporosis, obesity, diabetes, and gastrointestinal issues. A comprehensive token economy that utilized calories as tokens served as a foundation for providing reinforcement for behavior reduction and skill acquisition. Calories were then used as currency to purchase additional food items.

Diet and Weight Management

The weight management aspect of the program consisted of the following: calorie-controlled diet, skills training in meal planning and cooking, and an exercise regime. Daily calorie allotments and exercise regimes were prescribed and monitored annually by a physician expert in PWS. Individuals were trained in meal planning skills, including calorie computation, meal preparation, and calorie counting. Every individual was provided with a calorie sheet that included an individual's base allotment for calories per meal, prompts for dietary recommendations, and a section for tracking calories earned within the token economy.

Extra calories were offered as a reinforcer for exercise completion and demonstrating other targeted adaptive skills as well as the absence of problem behavior. These were added to an individual's "bank" of incentive calories, which functioned similarly to a bank ledger, and each person was able to apply these calories to meals as they saw fit. Once calories were used for food items, they were subtracted from the incentive calorie bank, and remaining calories were stored in the calorie bank to be used in a similar fashion at the next meal. Calories had no expiration date; however, a limit was placed by each individual's physician on the amount of incentive calories that could be used per meal to prevent dangerous food consumption.

Meal planning was trained as the main replacement behavior for food stealing. Giving individuals the opportunity to appropriately access food items, as well as to exercise choice and control over their diet was hypothesized to compete with food theft. Individuals were trained in meal planning immediately upon entering the program. Training included verbal instructions on using a calculator, and on completing each aspect of the calorie sheet and incentive calorie bank. Individuals were also provided training on how to identify and calculate caloric amounts. A calorie list was provided to each individual, which outlined the calories per serving size of each item available at the home. In addition, individuals were trained to calculate the calories of meals based on nutritional information provided by restaurants. These skills were necessary to assist individuals in preparing for less restrictive settings where they could plan and prepare their own meals. Instruction was also provided on using nutritional labels to identify calories per serving size. Individuals who had mastered meal planning and were eligible for transition to less restrictive environments were taught to create and calculate the calories of recipes they were interested in cooking. This generalized the skills learned during meal planning and calorie calculation and built upon this mastery to teach the measurement and conversion skills necessary for creating and carrying out calorically accurate recipes.

Behavior Management

Each individual was given an incentive sheet that served to track occurrences of both appropriate behavior and problem behavior and to track incentive calories earned throughout the day. At the end of each day, the sum of calories earned was added to the calorie bank. The occurrence or non-occurrence of a problem behavior was scored according to 2-h interval time segments which did not re-set if there was an occurrence of problem behavior. Calories were earned for each 2-h interval where targeted behaviors did not occur. Additional calories were earned based on the occurrence of adaptive behavior. This served as the foundation for reinforcing the absence of problem behaviors, and the occurrence of appropriate behaviors such as exercise, participating in activities, and meal planning.

Episodes of food stealing resulted in a requirement to pay back calories consumed, plus a 25-cal fine from the incentive calorie bank. For example, if an individual stole and consumed a 100-cal bag of chips, they were required to repay the 100 cal consumed, as well an additional 25-cal fine for a total of 125 cal. Episodes occurring close to mealtimes resulted in a reduction of food provided at that meal to prevent overeating per the discretion of the nursing department. If unpaid calories remained after these steps, these calories were forgiven such that no calorie deficit was incurred.

The AArc's foundational protocol included an "appropriate alternative to tantrums" procedure aimed at intervening early in a chain of problem behavior to teach de-escalation strategies and differentially reinforce appropriate alternative responses. A functionally equivalent response was identified by the individual's behavior analyst, and the staff and individual were trained in this behavior. When a problem behavior occurred, the staff implementing the procedure prompted an appropriate alternative based on behavior functions identified

in the individual's behavior plan. These alternative behaviors included requesting breaks, attention, and assistance, or other forms of problem solving. If an individual had success engaging in the alternative behavior, staff provided reinforcement in the form of praise, and at times calorie bonuses. In the case of escalation, a time-out procedure in a quiet, safe area was used in order to increase the safety of the individual and others, as well as to reduce attention provided to the problem behavior by an audience. During this time-out, individuals were prompted to self-time 5 min, and they had the option to engage in a solitary activity, such as coloring, or completing breathing exercises for the duration of the 5 min. Extinction/planned ignoring and monitoring were used to ensure the individual's safety but minimize the likelihood of reinforcing the behavior problem with attention.

If the individual was still engaging in any tantrum behavior after 5 min, the procedure was repeated. Once the individual completed the session and had not engaged in tantrum behaviors for 5 min, verbal praise was given, and the individual was prompted to either engage in the functionally equivalent communication response or return to scheduled activities. Under the DRO system, individuals were unable to earn incentive calories for intervals in which they demonstrated targeted behaviors. However, if an individual engaged in an appropriate alternative to a tantrum when prompted and did not escalate to a major problem behavior, they remained eligible to earn the DRO incentive for that interval, assuming they had not already engaged in a behavior problem that would prevent this.

Measures

To gain an understanding of the effects of this program on the weight and problem behavior of individuals with PWS, archival data from 45 individuals were examined. Data were selected from two points in time (2013/2014 and 2018/2019) to determine the long-term effects of the program on weight, food stealing, and other behaviors common for individuals with PWS.

Data were collected on outcome variables including weight, exercise, and behavioral topographies. Weight was measured on a standardized scale across the program and rounded to the nearest whole pound. Exercise was measured as a percentage of opportunity (POA), with each day representing one opportunity. Behavioral topographies were each recorded as frequencies, and each topography was operationally defined per the original program.

In order to accurately capture all topographies of food stealing for programmatic clarity, the AArc broke this behavioral class down into four separate operational definitions. Unauthorized food possession was defined as consuming food that had not been documented on a calorie sheet and the caloric value of the food item had not been subtracted from the available caloric balance. Inappropriate food consumption was defined as eating food that was not considered clean, healthy, and/or sanitary by commonly accepted community standards. Entering a food-restricted area was defined as entering an area that had been designated a food storage area, or that could reasonably be expected to contain food, without permission. Calorie sheet manipulation was defined as changing a calorie sheet, an incentive sheet, or any other document after it has been checked by staff, which then resulted in access to extra food. For the purpose of data analyses all food stealing topographies were aggregated into a single topography.

The response class of tantrum behavior was broken down into the following topographies. Vocal outburst (VOB) was defined as continued screaming, yelling, and shouting after 1 min of not responding to staff's attempts at redirection. Aggression was defined as hitting, kicking, punching, scratching, grabbing, throwing items, or using weapons that caused or may have caused physical harm to another individual. Property destruction (PD) was defined as any physical action that caused or may have caused damage to property. Elopement was defined as exiting or attempting to exit a facility, building, vehicle, or area in the community where supervision was being provided without staff permission.

Other behavioral topographies were defined as follows. Skin picking was defined as the use of fingers, fingernails, hands, or objects to cause damage to one's own skin. Self-injury was defined as using any items, or body part to cause physical harm to any part of one's own body. Rectal digging was defined as any contact between fingers and anus/rectum separate from normal toilet hygiene. Coprophagia was defined as any ingestion of feces. Rumination was defined as the regurgitation of previously swallowed food followed by rechewing and swallowing. Theft was defined as the act of taking an object belonging to another individual, as may be evidenced by being in possession of an object or objects owned by other individuals.

Data Analyses

Initial Data Collection

Data obtained during archival file review were originally collected by intake specialists and trained direct care staff. During the initial intake process, demographic information on date of birth, competency status, ethnicity, family involvement, diagnoses, and current medications were collected via interviews and file review. These data were updated as needed based on status changes and reviewed annually for accuracy. The training protocol for direct care staff responsible for collecting weight and behavioral data was as follows. Staff members assigned to supervise an individual were trained in the PWS protocol and any specific contingencies related to individual plans. Each staff member collected data on the frequency of

behavior problems throughout the day. Individual weights were taken each morning prior to breakfast on scales standardized across group homes and recorded on an incentive sheet. Clients were weighed on the same scale each morning to enhance reliability. Exercise was scored according to percentage of opportunities, with the assigned daily exercise routine counting as opportunity to exercise. Data were then manually entered by behavior services staff into an Excel spreadsheet for each individual. This information was collected to track each individual's clinical progress, and not for research purposes. Excel spreadsheets were stored on the program's server.

Direct care staff completed an initial 20-h behavior training upon hire, with a 4-h recertification course completed annually. Role play and testing were used to ensure competency before staff began supervising individuals or collecting data. Additionally, staff members attended bi-monthly meetings during which they were tested on knowledge of the program and where continued review and training on the behavior protocol were completed. Staff who did not attain 100% on a quiz received additional training until competency was achieved. Behavior services staff completed direct observation weekly of program intervention across settings to ensure fidelity of direct care staff and provided error correction training for any implementation or data collection mistakes.

Archival Data Collection

Program files were reviewed to identify individuals who had been enrolled in The AArc's PWS program for at least six continuous years at the time of this study. Data on age, gender, ethnicity, time in care, psychiatric medication, competency, and family involvement were collected from the agency's online database that holds all client demographic information. Age was calculated based on the listed date of birth. Time in care was calculated based on the listed admission date. Competency status was scored as either competent, competent with a guardian advocate, or incompetent according to the legal designation of the individual. Competent indicated the individual was able to make all decisions about their care independently, and guardian advocacy indicated the individual was court assigned an advocate to assist in decision making. Incompetent meant the individual required full oversight by a court-designated individual. Family involvement was scored "yes" if an active family contact was listed in the file and "no" if no family contact was present. The use of psychiatric medication was scored a yes if at least one psychiatric medication was prescribed according to the most recent psychiatric nursing note. These data were then cross-checked with the hard-copy client demographics files to ensure accuracy.

As data on weight and behavioral topographies were already entered into Excel files for each individual, the data collection procedure included moving the data to a new spreadsheet to ensure all personally identifiable information was removed. Excel functions were used to aggregate data for each individual and topography into 12-month time periods: August of 2013 to July 2014 and August 2018 to July 2019.

Analyses

The data were analyzed in four phases. First, descriptive analyses were conducted to provide a summary of the characteristics of the individuals. Second, statistical analyses and two-tailed paired sample *T* tests were used to evaluate the change in weight between the first and sixth year. Cohen's d effect size was also calculated to determine the effect size for weight loss across time. Third, behavioral topographies were analyzed using statistical analyses and a Wilcoxon's matched pairs test. The first and sixth years were compared for weight, food stealing, tantrum behavior, skin picking, and self-injury to analyze long-term outcomes. For each Wilcoxon's test, an effect size was calculated (Pallant 2007). Finally, theft, rectal digging, coprophagia, and rumination were not included in statistical analyses due to small sample size; however, the clinical significance of changes in these topographies was evaluated using percent reduction.

Results

Table 1 presents basic demographic information for individuals who were in the PWS program for at least 6 years at the time of data analysis. The majority of the population (93%) were Caucasian and between the ages of 30 and 50, with just over half female. Most individuals were legally competent (67%) and taking at least one psychiatric medication (84%). The average time in the program was just over 15 years ($M =$ 15.4 years). Table 2 displays weight data for years one and six. Individuals lost an average of 12 pounds between years one and six, for an overall average of two pounds per year. The range of weight gain and loss varies greatly from 41 pounds gained to 86 pounds lost over a 6-year period. Weight gain was often attributed to home visits, but those data were not included in this study. The results of the paired-sample *t* test indicate that there was a significant effect for weight between the first and sixth years ($t(44) = 3.46$, $p < .001$, $d =$ 0.52), with individuals weighing less in the sixth year than year one.

Table 3 presents data on the occurrence of each response topography of problem behavior in the sample population. A topography was marked as occurring if the individual was recorded as engaging in the behavior at least once within the data set, at any point during the 6-year period. The following behaviors were reported for at least one individual in the program: food stealing, VOB, aggression, PD, skin picking,

Table 1 Demographic characteristics of the individuals with Prader–Willi syndrome

Variable	Number	Percentage	Mean (SD)	Range
Age	45		*M* = 40.15 (9.8)	20–57
Gender	45			
Female		53% (*n* = 24)		
Male		47% (*n* = 21)		
Ethnicity	45			
Caucasian		93% (*n* = 42)		
Hispanic		4% (n = 2)		
African American		2%. (*n* = 1)		
Time in care	45		*M* = 15.4 (5.9)	6–28
Psychiatric medication	45			
Yes		84% (*n* = 38)		
No		16% (*n* = 7)		
Competency	45			
Competent		67% (*n* = 30)		
Competent with guardian advocacy		20%. (*n* = 9)		
Incompetent		13% (*n* = 6)		
Family involvement	45			
		96% (*n* = 43)		
		4%. (n = 2)		

elopement, self-injury, theft, rectal digging, coprophagia, and rumination. Other behaviors that were observed but not formally tracked included hoarding, repetitive questioning, and arguing. Food stealing was reported for 98% of individuals, with behaviors included in the response class of tantrums (VOB, aggression, PD, elopement), and skin picking occurring in more than 50% of the population.

The results of the Wilcoxon's matched pairs analyses and effect sizes for each comparison are displayed in Table 4. All analyses showed statistical significance, with year six being associated with lower rates of targeted behaviors. As mentioned, theft, rectal digging, coprophagia and rumination were not included in statistical analyses due to small sample sizes; however, all four behaviors show a clinically significant reduction across time in treatment. Rectal digging decreased 92% from year one to year six, rumination decreased 89%, coprophagia decreased 49%, and theft decreased 58%.

Table 2 Weight data

Weight	Mean (SD)	Range
Year 1	156 (35)	107–262
Year 6	144 (23)	99–199
Weight lost Years 1–6	12 (24)	− 41—86

Weight in pounds at time periods 1 and 6, and weight loss during the 6-year span, reported in mean, standard deviation, and range. Negative numbers indicate weight gained

Discussion

To date, few interventions have been demonstrated to be comprehensively effective in managing the variety of physical and behavioral challenges associated with PWS. It is therefore encouraging that the treatment package implemented by The AArc shows strong historical evidence to suggest efficacy in the long-term management of weight, food-stealing, and behavior problems. The data presented indicate that during their time in The AArc's program, individuals demonstrated

Table 3 Prevalence of behavioral topographies among the individuals with Prader–Willi syndrome (*n* = 45)

Variable	Number (%)
Food stealing	44 (98%)
Vocal outburst	41 (91%)
Aggression	33 (73%)
Property destruction	32 (71%)
Skin picking	26 (58%)
Elopement	23 (51%)
Self-injury	12 (27%)
Theft	11 (24%)
Rectal digging	3 (7%)
Coprophagia	2 (4%)
Rumination	2 (4%)

Number of individuals who engaged in each topography at least once during the 6-year duration of data collected

Table 4 Data analyses of behavioral topographies

	Year 1 *M* (SD)	Year 6 *M* (SD)	Total *M* (SD)	*Z* Years 1–6	*d* Years 1–6
Food stealing	81.9 (118.6)	31.3 (86.2)	322.8 (662.5)	−4.191***	0.62
Vocal outburst	153.2 (177.6)	55.3 (74.9)	553.6 (751.1)	−3.938***	0.59
Aggression	25.5 (34.3)	8.2 (12.0)	99.8 (154.5)	−4.279***	0.64
Property destruction	17.7 (27.0)	5.7 (8.3)	58.6 (92.0)	−3.515***	0.52
Skin picking	83.3 (327.3)	17.3 (44.1)	284.0 (1057.1)	−2.631**	0.39
Elopement	10.6 (20.5)	2.2 (4.7)	30.7 (55.3)	−3.862***	0.58
Self-injury	5.4 (14.7)	1.7 (10.7)	20.0 (97.3)	−2.524*	0.38
Theft	2.4 (11.1)	1.0 (3.2)	8.6 (27.0)		
Rectal digging	12.5 (73.6)	1.0 (6.1)	17.5 (92.9)		
Coprophagia	1.7 (7.9)	0.8 (4.7)	7.9 (38.8)		
Rumination	3.6 (17.9)	0.4 (1.9)	5.7 (27.0)		

Results of Wilconox's matched pairs tests and effect size calculations for targeted behaviors. Wilconox's matched pairs tests were not conducted for theft, rectal digging, coprophagia, and rumination due to small sample sizes

*Significant differences between years at $p < .05$

**Significant differences between years at $p < .01$

***Significant differences between years at $p < .001$

statistically significant decreases in weight and food stealing, as well as in all behaviors classified as tantrum behaviors (VOB, aggression, PD, elopement). Previous behavior analytic research on weight and food stealing management has suggested that discrimination training, exercise, self-monitoring, token economies, response-cost, and DRO and differential reinforcement of alternative behavior are all effective components of treatment packages for managing weight and food stealing within PWS (Altman et al. 1978; Maglieri et al. 2000, Page et al. 1983, b; Rone 2010; Singh et al. 2008, 2011). All of these strategies are included as aspects of The AArc's treatment program, suggesting that these methods may be effective as part of a long-term treatment package.

All aspects of the protocol are consistent with evidence-based practice on reducing problem behaviors in other populations. Specifically, teaching a functionally equivalent response while implementing extinction for the problem behavior has been demonstrated to be highly effective with adults with developmental disabilities, even when problem behavior is intermittently reinforced, as can occur with dangerous behavior that cannot be fully ignored (Chezan et al. 2014; Shukla and Albin 1996; Worsdell and Iwata 2000). Time-out has been extensively researched as a method for reducing inappropriate behavior (Donaldson and Vollmer 2011). In this protocol it may serve additional purposes including removing the individual from aversive stimuli likely to escalate the behavior, and reducing attention from bystanders, therefore reducing the overall magnitude of reinforcement for behaviors maintained even partially by attention. Despite consistencies with evidence-based practice, specific experimental replication is necessary to truly evaluate the effects of these methods on behavior problems associated with PWS.

The presence of clinically significant decreases in behaviors assumed to be automatic in function (rectal digging, ruminations, and coprophagia) indicates that these behaviors may respond well to behavioral strategies, despite the fact that behavior maintained by automatic reinforcement is the most difficult to treat behaviorally (Vollmer 1994). These results are supported by previous research targeting rectal digging for individuals with PWS which indicated that antecedent control (limited bathroom use), functional communication training, and DRO can effectively manage this behavior (Stokes and Luiselli 2009). The extremely low prevalence (4–7%) of rectal digging, rumination, and coprophagia among individuals at The AArc compared with the 15% prevalence reported in research (Hanchett 1994) may suggest that these behaviors interact significantly with environmental variables such as diet, medical intervention, and social contingencies.

Limitations and Future Research

As mentioned, the use of archival data presents with significant limitations. The reductions in weight and problem behavior cannot be directly attributed to specific aspects of the program. A further limitation is the failure to collect data on body mass index (BMI) as a measure of appropriate weight. This omission prevents the ability to report on the relation of weight to obesity-related disease risk. Data were not reported on whether or not individuals were already in their targeted weight range or met their targeted weight range during the

period of data collection. Individuals in this study were prevented from dropping below their targeted weight range by the addition of calories by their overseeing physicians. The annual rate of weight loss of the population may have been understated if a portion of the population were maintaining rather than losing weight.

Although the data suggests that The AArc's program is effective as a package, without further investigation it is impossible to determine which aspects of the treatment program caused the behavior change, and if any strategies may be equally as effective if used as a sole intervention. The lack of experimental manipulation means that the presence of environmental variables cannot be accounted for. The effect that aspects of being in a residential facility (e.g., increased supervision, modeling by peers) or other potentially significant aspects of this environment cannot be quantified in this type of study. Experimental replication and component analyses of treatment variables are needed to definitively state the efficacy within the PWS population.

Treatment integrity is similarly difficult to evaluate. Given that data were originally collected as part of a residential treatment program, the extent to which treatment components were implemented consistently is difficult to quantify. Treatment within residential programs is subject to staff turnover, changes in individual behavior plans, caregiver intervention, individual medication changes, and many other variables that interact with programmatic protocols. Needless to say, implementation is unlikely to have held up to experimental standards. Previous research indicates that low treatment integrity may result in reduced treatment gains, including failure to acquire skills, or delays in mastery (Brand et al. 2019). That being said, the fact that this treatment program was effective despite imperfect implementation may make it of higher interest to residential facilities.

Further research is needed to validate and compare treatment components in an effort to quantify efficacy and recommend improvement treatment packages such as that implemented by The AArc. There is still no best practice for treating PWS, likely as a result of its rare and complicated nature. Additional studies that evaluate treatments from different fields and disciplines both individually and in conjunction are necessary to help improve quality of life and long-term treatment outcomes. Given how complex PWS is both medically and behaviorally, multi-disciplinary approaches warrant investigation.

Authors' Contributions KB: designed and executed the study, completed the data collection and analyses, and wrote the paper. AG: collaborated with the design of the study, data analyses, and editing of the manuscript. ML: contributed to the original authorship of the program's protocol and collaborated with the editing of the final manuscript. MS: collaborated with the editing of the final manuscript. All authors approved the final version of the manuscript for submission.

Data Availability Data are unavailable due to the small sample size from an identified site which could result in loss of confidentiality.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

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