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A measure of food seeking in individuals with Prader–Willi syndrome

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Abstract

Background—Individuals with Prader–Willi syndrome (PWS), a chromosome 15 genetic disorder, often have a significant preoccupation with food and problem behaviour related to food seeking is often prevalent.

Methods—In the present study, we compared how individuals with PWS responded on a survey regarding the acceptability of food in various locations that varied according to degree of appropriateness for human consumption (e.g. food on a plate, food in a garbage can). For a subgroup of participants, we observed how they actually responded when placed in a room with food items placed in the same locations depicted in the survey. In the first part of the study, three groups (25 typically developing individuals, 7 individuals with intellectual disability (ID), and 19 individuals with PWS) responded to a visual survey to determine the degree of acceptability of food items in various locations (e.g. on a table near a hairbrush, on the floor behind a toy box, in a trash can). In the second part of the study, these food items (popcorn, jelly beans) were placed in the 12 locations described above. Nine individuals diagnosed with PWS (deletion type) and three individuals with ID were given some break time in the room for 15 min. The amount of food consumed, the time spent food seeking, and time spent interacting with materials were measured.

Results—Results of the survey indicated that the PWS group differed significantly with regard to how they responded on the survey from the typically developing group, but did not differ significantly from the ID group. Results of the food seeking observations indicated that only three individuals with PWS ate a significant number of items. The three individuals did not differ from the rest of the group according to IQ or compulsivity score; however, they had significantly lower body mass index (BMI) scores and were younger than the other participants.

Conclusions—The findings from the survey indicate that individuals with PWS are able to discriminate the appropriateness of eating items in more or less contaminated areas; however, the amount of time spent seeking food and the amount of food covertly consumed appeared to depend more directly on age and BMI.

Keywords

food seeking; food stealing; Prader-Willi syndrome

Introduction

Prader-Willi syndrome (PWS) is characterized by hypotonia, hypogonadism, hyperphagia, early childhood obesity, small hands and feet, short stature, learning and behaviour problems, and a characteristic facial appearance (Butler 1990). Prader-Willi syndrome is considered the most common genetic cause of life-threatening obesity. A chromosome 15 q11-q13 deletion is seen in about 70% of individuals, while the remaining individuals have maternal (uniparental) disomy 15 or imprinting defects (Butler & Thompson 2000). This intense preoccupation with food, lack of satiation, and food seeking are some of the most striking characteristics of the disorder (Dykens *et al.* 1996,1999; Dykens & Kasari 1997; Dimitropoulos *et al.* 2002). If left unaddressed, the excessive eating may lead to marked obesity and other health complications. Caregivers often resort to restrictive practices such as locking away food and closely supervising individuals in the community so that covert food stealing and overeating do not occur (Butler & Thompson 2000).

Although the exact mechanism causing hyperphagia is unknown, it is clear that individuals with PWS have a higher threshold for satiety than typical populations (Holland *et al.* 1995). Efforts to quantify food-motivation have been limited given that the behaviour is often covert and that dietary restrictions make experimental manipulations with excessive food consumption or food stealing difficult to measure. In a study by Zipf & Berntson (1987), the authors found that some participants with PWS consumed up to 125 quarter sandwiches in a single sitting, and had to be stopped from eating larger amounts. Holland *et al.* (1995) evaluated the amount of overeating and degree of satiety in 13 participants with PWS. They allowed the participants to have free access to food and measured the degree to which they felt hungry before, during, and after the free access meal. The mean number of calories consumed by the PWS group was 1292 compared with 369 calories by control participants. In addition, the PWS group reported increased feelings of hunger within 20 min after the free access meal, whereas control participants reported that they weren't hungry up to 100 min after eating.

Additionally, it was once thought that people with PWS will simply eat anything regardless of amount or flavour (Pipes & Holm 1973), but there is empirical evidence that individuals with PWS have food preferences that are similar to those without PWS. For example, Taylor & Caldwell (1985) demonstrated that people with PWS have clear preferences for sweet to salty, sour, and bland foods and will choose the more preferred foods over others. Rankin & Mattes (1996) also demonstrated that foods with a high degree of fat content were almost equally preferred to sweet foods by individuals with PWS. Control participants tended to prefer high-fat foods slightly to sweet foods. Fieldstone *et al.* (1997) demonstrated that individuals with PWS had a greater preference for high-carbohydrate foods than obese and normal weight control participants. These studies indicate that individuals with PWS appear to have similar but not identical food preferences to individuals without PWS and that excessive food-seeking is not related to an inability to discriminate between pleasant and unpleasant foods.

Taylor & Caldwell (1985) also assessed whether given the choice between a small amount of highly preferred vs. a large amount of less preferred food, individuals with PWS were more likely to select the small amount of preferred food. Glover *et al.* (1996) extended this research by including a control group of individuals with intellectual disabilities (ID) but without PWS. They found that participants with PWS selected a small amount of preferred items only over a large amount of their least-preferred items. Finally, Joseph *et al.* 2002 found that individuals

with PWS were more likely to choose a larger quantity of food delivery regardless of the amount of delay and degree of preference of the food item. These findings indicate that individuals with PWS may vary significantly in their choice making, depending on the quality and quantity of the food, as well as the length of the delay to access.

One area in need of further study is the degree to which individuals with PWS engage in inappropriate food consumption (including stealing food, eating contaminated or inedible foods, or eating unusual food combinations). Dykens (2000) surveyed 50 individuals with PWS and determined the degree to which they would be willing to eat contaminated (e.g. cake with a bug in it), somewhat contaminated (cake on a clean sponge), and unusual food combinations (e.g. ice cream on steak). Many more individuals with PWS indicated that they would be willing to eat contaminated, somewhat contaminated, and highly unusual combinations of food than IQ matched peers or typically developing (TYP) participants, even though they understood that it was not considered acceptable to eat these foods.

Dykens' (2000) study demonstrated that individuals with PWS can provide verbal report about what is and isn't acceptable to eat. It also suggests that when the opportunity to consume food arises, the behaviour of individuals with PWS may not correlate with their verbal report. Thus, it is not clear how an individual with PWS would behave given the opportunity to eat food items that varied by level of contamination or acceptability. There have been several studies evaluating pica, a similar problematic eating disorder involving the ingestion of inedible objects, which may provide a methodology for evaluating covert eating in individuals with PWS. For example, Donnelly & Olczak (1990) and Piazza *et al.* 1996 baited a room with mock cigarette butts that did not contain nicotine to evaluate the frequency of cigarette pica under controlled experimental conditions. Additionally, Piazza *et al.* (1998) used this procedure with several topographies of other forms of pica to evaluate the circumstances under which pica was more likely to occur (e.g. alone, with others present, during tasks). These studies provide a methodology that may be useful for studying covert food seeking (and eating) in individuals with PWS.

The purpose of the current study was to evaluate food-seeking behaviour in individuals with PWS in two ways. First, a visual survey was conducted with three groups: (1) individuals with PWS, (2) individuals with ID, and (3) TYP individuals to evaluate willingness to eat food in a variety of locations in a room. The responses of the TYP group were compared with the other two groups. Second, individuals with PWS and a control group of individuals with ID were situated in a room with small amounts of food (in the same locations pictured in the survey) to determine if there were differences between the two groups in the amount of time spent food seeking, the number and location of items eaten.

Study 1: food location survey

Method

Participants and setting—Participants were 25 TYP children and adults, seven individuals with ID, and 19 individuals with PWS. The participants were part of a larger study group comparing food seeking and other compulsive behaviours in individuals with PWS and control groups. Participant assessments were conducted over a 1–2-day period. See Table 1 for a description of demographic characteristics for each group of participants.

Twelve digital photos of three pieces of popcorn and one jelly bean in various sites in a behavioural testing room were used for the study. The photos were presented in pairs so that each one was compared with all others, resulting in 66 total pairs of photographs (see Fig. 1 for an example). The photo pairs were shown to each participant using a laptop or desktop computer. The order of pairs was randomized along with location on the screen (i.e. right or

left side). Participants were asked to choose the photograph (either by pointing with his/her finger or with the cursor) that pictured what he/she would be 'more willing to eat'. An experimenter recorded on a data sheet which photo in the pair was selected. The session took approximately 15 min to complete.

All 12 of the pictures were classified as least acceptable (worst), moderately acceptable (better), or most acceptable (best) to eat based on the responses by the TYP group (see Table 2 for a list of pictures in each category). Using the TYP group as the criterion, each participant's responses in the PWS and the ID groups were categorized according to these acceptability groupings.

Results

All statistical analyses were performed using SPSS version 11.5. A significance level of 0.05 was chosen prior to the study. Mann–Whitney *U*-tests were used to determine if there was a difference between each group according to the classification scheme described above (i.e. better vs. worst, best vs. worst, best vs. better). The Mann–Whitney *U*-test was chosen because of the relatively small sample sizes in each of the three groups. Results of the study showed that the PWS and ID groups differed significantly from the responses made by the TYP group and there was no statistically significant difference in the responses between the PWS and ID groups.

Study 2: food seeking sessions

Method

Participants and setting—Nine individuals (four females and five males) with PWS participated in the study. The mean age of the PWS participants was 18 years of age. Three individuals with ID (two female, one male) participated as controls. The mean age of the control group was 13.5 years of age. Sessions were conducted in the same behavioural testing room that was used to make the digital photos described in Study 1. Hidden cameras were placed in the far corners on opposite sides of the ceiling so that the participant could be observed at all times in any location in the room. Consent for participation and videotaping was provided for the study by the parents or legal guardians of the participant.

Procedures—The room was set up with the popcorn and jelly beans in the 12 sites exactly as they were depicted in the survey in Study 1. Each cluster of food items was placed on a clean piece of transparency film and then placed in each location in the room. All areas of the room were cleaned prior to the session and containers used in the session (e.g. wastebasket) were brand new and used only for this study. Sessions were conducted immediately before lunch to maximize the likelihood that participants were hungry. Prior to the session, participants were asked to select a low-calorie snack to eat during their break and were shown where toys, puzzles, magazines and games were located in the session room. Participants were then told that his or her caregiver was down the hall completing rating scales and that the participant would be undisturbed for the next 15 min. The research staff placed a kitchen timer in the room, set for 15 min, and left room. When the timer went off, the research staff reentered the room and escorted the participant to the next activity.

Response definitions—The 15-min sessions were videotaped and scored for several behaviours including: food seeking (moving around room, going through items on table or in toy box, looking under furniture, going through wastebasket), handling of food (if participant manipulated or touched the popcorn and/or jelly beans prior to or after placing it in his or her mouth), consumption of appropriate snack food, consumption of the target food items, and time spent interacting with the leisure items.

Data collection—Throughout the 15-min session, caregivers and research staff observed the participant via the hidden cameras on a TV monitor in the next room. A video cassette recorder recorded the sessions. Sessions were subsequently scored independently by two trained observers. Interobserver agreement was collected for 54% of the sessions (for 7 out of 13 participants). The session time was divided into consecutive 10-s intervals and a partial interval data collection system was used. Agreement was defined as both observers recording the behaviour within the same 10-s interval and was calculated by comparing the number of agreements divided by the number of agreements plus disagreements for each behaviour. Overall agreement for all behaviours across all participants was 97.9% (range 93–100%).

Results

Table 3 shows the mean number of intervals that PWS and control participants spent seeking food, playing with food, consuming food, and playing with/interacting with materials in the room. In addition, the mean percentage of popcorn and jelly beans consumed by each PWS and control participant is also indicated. On average, only 18% of the food items were eaten. This was primarily attributed to three participants who ate a significant proportion of food (consumers) compared with the other participants (non-consumers). Four individuals never touched the food or spent any time looking for it. Consumers and non-consumers were compared according to age, IQ, and body mass index (BMI) using a *t*-test with $P = 0.05$. The two groups varied significantly on age (consumer group mean age = 11.7 years; non-consumer group mean age = 24 years) and BMI (consumer group mean BMI = 24; non-consumer group mean BMI = 40.5).

Discussion

Results of the food seeking survey showed that the PWS and ID groups gave responses that differed significantly from the TYP group on the food survey. Possible differences on the survey results may be because of general ID rather than PWS, specifically because both the PWS and the ID group differed from the TYP group on the survey. Inconsistencies in responding on the survey may have been due to confusion with the survey or inability to adequately communicate responses that were related more to the individual's intellectual level rather than their actual food preference.

Interestingly, individuals with PWS were not more likely to engage in covert food seeking or consumption when placed in a room with reported preferred food items in the baited room study. This was surprising given that for all participants, parents reported that food stealing was a problem. Possible explanations for why more participants with PWS did not eat target items during the session include: (1) the setting was too artificial; (2) reactivity to the study situation; (3) the participants were afraid that caregivers would catch them eating (even though we tried to reassure them that the caregivers would not); or (4) they saw the cameras and knew they were being observed. Anecdotal evidence leads us to suggest that (1) and (2) are most likely explanations for this particular study group. For the three individuals with PWS who did eat a moderate percentage of food (the consumers), they may have been more likely to eat the snack foods because all three were on extremely restricted diets.

As a result of these mixed findings and the differences between the groups, the current study is rather limited in the conclusions that can be made; however, our findings do suggest that future research in this area warrants an analysis of this behaviour using more naturalistic conditions and observers. For example, conducting the assessment at the individual's home would help reduce reactivity to the setting and situation, but may also make experimental control and observation more difficult. In addition, future research might include the use of highly preferred food items for each individual rather than items chosen by the experimenter. The number of sites used should be decreased given that the density of food in the small session

room may have made our participants more suspicious of the situation. Finally, given evidence of behavioural differences between genetic subtypes, it will be interesting to compare subtypes on this task to see if individuals with the deletion were more or less likely to engage in food seeking and consumption compared with individuals with maternal disomy.

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Figure 1.
An example of a digital photo set that was used in the survey in Study 1.

Table 1
Demographic information for Study 1 participants

	PWS (N= 19)	TYP group (N= 25)	ID group (N= 7)
Subtype			
Deletion	12	N/A	N/A
Maternal disomy	5		
Other/Unknown	2		
Gender			
Male	7	7	4
Female	12	15	3
Unknown	N/A	3	N/A
Age 1–12	4	4	4
Age 13–17	3	0	3
Age 18–25	7	1	0
Age 26–30	2	7	0
Age 31–40	2	4	0
Age 41–50	1	6	0
Age 50 and over	0	3	0

PWS: Prader-Willi syndrome; TYP: typically developing; ID: intellectual disability; N/A: not available.

Table 2
Food photographs characterized as worst, better, and best

Worst	In trash can On floor by spider On floor behind toy box On table next to crumpled tissue
Better	On table next to hairbrush On edge of bulletin board attached to the wall On the seat of a chair
Best	On table next to pill (medication) container On table behind a plastic container On table next to tissue box On table On paper plate

Table 3

The percentage of intervals spent food seeking, handling food, consuming food, playing with leisure items, and the mean percentage of items consumed by study group

	Food seeking	Food handling	Consuming food	Playing	Total number of items eaten
PWS total	16	0.2	1	60.9	16.4
PWS consumers	41.5	0	3.1	34.2	47.2
PWS non-consumers	4.9	0.2	0.5	72.4	3.2
ID control	0	0	0.4	43	3.7

PWS: Prader-Willi syndrome; ID: intellectual disability.