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Perceptions of Body Image by Persons With Prader-Willi Syndrome and Their Parents

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Abstract

Prader-Willi syndrome is a genetic disorder characterized by obesity. The Figure Rating Scale (Stunkard, Sorensen, & Schulsinger, 1983) was completed by 43 individuals with this syndrome to determine their level of dissatisfaction with their body. Their parents also completed this scale regarding their child to determine whether they were dissatisfied with their child's body status. Results showed that individuals with Prader-Willi syndrome were dissatisfied with their body. Parents also were dissatisfied with their child's body. Results of this study demonstrate that the responses of persons with Prader-Willi syndrome on the Figure Rating Scale show significant discrepancies between how they think they look and how they wished they looked.

Prader-Willi syndrome, a genetic disorder that occurs in approximately 1 in 10,000 to 15,000 births, is characterized by marked obesity and specific behavioral and cognitive attributes (Butler, Theodoro, Bittel, & Donnelly, 2007). The genetic basis of Prader-Willi syndrome is a de novo paternal deletion of the long arm of chromosome 15 (15q11–13) in about 70% of cases, maternal disomy of chromosome 15 in 25% of cases, and an imprinting center mutation in the remaining 1% (Mascari et al., 1992; Nicholls, 1993). There are two manifestations of the deletion subtype in Prader-Willi syndrome: the Type I deletion spans a larger area of the critical region of chromosome 15 than does the Type II deletion (Butler, Bittel, Kibiryeve, Talebizadeh, & Thompson, 2004; Milner et al., 2005).

Prader-Willi syndrome is the most commonly known genetic cause of obesity in humans, with obesity and hyperphagia (excessive appetite) developing early in life and resulting in an individual's total body fat being, on average, three times higher than that of the general population. Persons with Prader-Willi syndrome often weigh more than 200% of their ideal body weight, which can lead to many weight-related health problems, such as sleep apnea, osteoporosis, impaired respiratory function and hypoventilation, reduced physical activity, and endocrine dysfunction (Butler et al., 2007). These health problems are similar to those of

persons who are obese and do not have Prader-Willi syndrome, but it is not known if persons with the syndrome experience the same psychological concerns around being obese as individuals who do not have Prader-Willi syndrome.

For individuals with obesity and other eating disorders, issues associated with poor body image and body dissatisfaction have been well-established. One measure of body image is the Figure Rating Scale (Stunkard et al., 1983). This measure has been widely used in body image research with a variety of populations, including women (e.g., Cash & Henry, 1995); between younger and older women (e.g., Bedford & Johnson, 2006); African American, Hispanic, and Caucasian men and women (e.g., Demarest & Allen, 2000), Mexican American women (e.g., Cachelin, Monreal, & Juarez, 2006); and women who have successfully undergone weight loss treatments (e.g., Foster, Wadden, & Vogt, 1997). It has even been used with children as young as 11 years (Sherman, Iacono, & Donnelly, 1995). There is also increasing evidence that having a positive body image is one of the most robust predictors of success in weight loss treatments (e.g., Plesa-Skwerer, Sullivan, Joffe, & Tager-Flusberg, 2004; Teixeira et al., 2002) and that overweight people are more dissatisfied with their body image than are persons who are closer to their ideal body mass index–BMI (Forbes et al., 2005).

Much of the research has shown that there is a tendency for women to select an ideal body size significantly thinner than their perceived current size (e.g., Cachelin, Striegel-Moore, & Elder, 1998; Cohn & Adler, 1992; Fingeret, Gleaves, & Pearson, 2004; Gleaves et al., 2000), indicating that many women are dissatisfied with their bodies regardless of their weight (Forbes et al., 2005). Males, on the other hand, tend to overestimate their body size (Sorbara & Geliebter, 2002). These gender differences in body image have been demonstrated to appear prior to puberty, with adolescent girls having greater body dissatisfaction than do adolescent boys (Mayville, Katz, Gipson, & Cabral, 1999).

Obese individuals tend to underestimate their body size, which is in contrast to individuals with eating disorders who are likely to overestimate their size (Bell, Kirkpatrick, & Rinn, 1986; Forbes et al., 2005; Holder & Keates, 2006). Given this information, researchers have investigated the accuracy with which a person might identify a picture (e.g., photo, silhouette) that most accurately reflects their current body size or BMI. Viner et al. (2006) found that the accuracy of weight perception decreased with increasing BMI in boys but that accuracy increased as girls' BMIs increased. These investigators also found that a surprisingly low percentage of overweight boys and girls recognized themselves as "too heavy," although girls were considerably more accurate than boys.

Although fairly well-studied in the general population (Chacelin et al., 2006), self-concept, particularly as it relates to body image and dissatisfaction, has not been widely examined in individuals with intellectual disabilities or in individuals with a known genetic marker that may directly affect body weight, such as Prader-Willi syndrome. In one of the few published studies, Weiss, Diamond, Denmark, and Lovald (2003) found that individuals with intellectual disabilities are more prone to developing negative self-concepts and low self-esteem, due to their perceived intellectual inadequacy, pervasive failures in academic and social domains, and stigmatization. Compared to their peers, adolescents with intellectual disabilities have poorer self-image and more feelings of frustration and inadequacy (Weiss et al., 2003). For individuals with Prader-Willi syndrome in particular, obesity and social difficulties can significantly impact self-concept (Plesa-Skwerer et al., 2004).

Our purpose in the current study was to examine body dissatisfaction in persons with Prader-Willi syndrome and how persons with Prader-Willi syndrome differ from established norms (Bulik et al., 2001) with regard to their body dissatisfaction. We also sought to determine whether persons with Prader-Willi syndrome were able to accurately identify their current body

status based on BMI. Because parents can be influential in their child's body perception, parent's dissatisfaction with their child's body and whether parents were accurate in reporting their child's body size also was examined. Additionally, we analyzed the interaction between the body dissatisfaction of persons with Prader-Willi syndrome across a variety of variables, including gender status, age, growth hormone status, and genetic subtype (i.e., deletion or uniparental disomy subtype of Prader-Willi syndrome).

Method

Participants

Forty-three individuals with Prader-Willi syndrome and 42 of their parents participated. The majority of this sample were participating in a larger study evaluating compulsive behavior and excessive eating in individuals with Prader-Willi syndrome (33 of the 43 individuals, or 77%). Participants were recruited locally, regionally, and nationally through the Prader-Willi Syndrome Association (USA) website and the Prader-Willi Syndrome Association annual conference. All individuals gave assent, and parents signed informed consent approved by institutional review boards.

BMI Classification

On the basis of height and weight, we calculated current BMI for each participant using the BMI calculator available on the Centers for Disease Control (CDC) website. The child and teen calculator was used for children (i.e., 19 and under) and the adult calculator, for adult participants. Categories of weight classes (e.g., obese, overweight) provided by the CDC were used to group participants (see Figure 1)

Genetic Classification

High-resolution chromosome analysis, fluorescence in situ hybridization, and DNA microsatellite analysis were used to confirm the deletion or maternal disomy genetic subtype status (Butler, Bittel, & Talebizadeh, 2002; Miralidhar & Butler, 1998).

Procedure

The Figure Rating Scale, a reliable and easy-to-administer self-report measure of body image, consists of nine pictures (silhouettes that are either male or female) presented to a participant. The pictures range from persons who are extremely thin (Silhouette 1) to extremely obese (Silhouette 9). A person is asked at least two questions: to identify the picture they "think they look" most like and the picture they "wish they looked" the most like (Thompson & Altabe, 1991). Once scored, three specific measures are obtained: current size (participant look), desired size (participant wish), and a body dissatisfaction score (look minus wish), which is the measure of body dissatisfaction (e.g., Altabe & Thompson, 1992; Fallon & Rozin, 1985). The Figure Rating Scale has been shown to have strong test-retest correlations (significant at the .001 level) for both questions regarding how a participant thinks they look (females, .89; males, .92) and how they wish they looked (females, .71; males, .82) (Thompson & Altabe, 1991).

All 43 individuals with Prader-Willi syndrome completed the Figure Rating Scale independently. One parent of each participant completed a second Figure Rating Scale regarding their perception of their child ($N = 42$). For one individual with Prader-Willi syndrome, no parent score was collected. A body dissatisfaction score greater than 0 indicates a desire to be thinner, and a score less than 0, a desire to be heavier (e.g., Fitsgibbon, Blackman, & Avellone, 2000). To determine the parents' body dissatisfaction score for their child, we also subtracted their wish score from their look score.

Must, Phillips, Stunkard, and Naumova (2002) identified a BMI score associated with each of the nine silhouettes on the Figure Rating Scale. These authors determined that the thinnest silhouette was a BMI of approximately 15, the middle silhouettes were overweight at BMIs approximately 30, and heaviest silhouettes were obese at BMIs over 30. To judge the participant's accuracy in choosing their current silhouette, each of them was assigned a silhouette that most closely matched their BMI based on Must et al. (2002) criteria (see BMI information in Table 1). Next, an accuracy score was determined by subtracting the silhouette that matched the participant's BMI from the silhouette chosen by the participant for how they thought they looked (participant look). The participant was considered accurate if the accuracy score was zero (e.g., Loh, Meyer, & Leckband, 2008). The accuracy of the parent's choice was defined in the same manner.

Data Analysis

Data were analyzed using SAS (version 9.1, SAS Institute Inc, Cary, NC). We conducted Wilcoxon signed-rank tests to examine the body dissatisfaction scores of persons with Prader-Willi syndrome and their parents. This test was also used to examine the degree of accuracy of body size by the person with Prader-Willi syndrome as well as their parent's accuracy.

We carried out bivariate analyses using *t* tests to test whether the body dissatisfaction and accuracy were associated with gender, genetic subtype, and growth hormone status. We performed correlation analyses to test whether body dissatisfaction and accuracy were correlated with demographic characteristics such as IQ, age, and BMI. Fisher's exact test was used to test whether accuracy scores of persons with Prader-Willi syndrome were associated with any of the above covariates.

Further, we used multiple regression analysis to identify and evaluate factors associated with body dissatisfaction. The final regression model was built by backward selection, with $p < .1$ for retention. The *p* values are two-sided and considered statistically significant at the .05 level. We also conducted regression analyses to determine whether accuracy score was predicted by any of the factors. The same analysis was applied to parents' Figure Rating Scale accuracy scores.

Finally, Bulick et al. (2001) reported normative data for individuals 18 years or older by gender and age group. To determine whether there was a difference between subjects with Prader-Willi syndrome and typical controls, the mean body dissatisfaction score of Prader-Willi syndrome subjects ($n = 27$) was tested against the reported mean body dissatisfaction score of typical subjects using one sample *t* test, stratified by age and gender.

Results

Demographic characteristics for the study sample are summarized in Table 1.

According to the Wilcoxon signed-rank tests, testing against the null hypothesis (i.e., that the median difference between look and wish is 0), participants with Prader-Willi syndrome experienced significant body dissatisfaction (see Table 2). Parents of the participants also experienced significant dissatisfaction with the body of their child (Table 2). We performed multiple regression analyses to investigate whether body dissatisfaction of persons with Prader-Willi syndrome and their parents are associated with any of the following factors: BMI, age, gender, subtype, growth hormone therapy status, and IQ. For persons with Prader-Willi syndrome, backward selection procedures identified that BMI and gender were significantly associated with body dissatisfaction. That is, the greater the BMI of persons with Prader-Willi syndrome, the greater their body dissatisfaction (see Table 3). Further, modeling of the interaction between BMI and gender was marginally significant, $p = .06$, with females tending

to experience greater dissatisfaction more quickly as BMI increased as compared to males (see Figure 2). The same method was applied to parent's perception of their child's body to identify any risk factors. Gender was the only variable found to be significant. Parents of males tended to have a greater body dissatisfaction score ($M = 3.07$, $SD = 1.94$) than parents of females ($M = 1.52$, $SD = 2.14$). Finally, Figure 3 shows the mean body dissatisfaction score of persons with Prader-Willi syndrome by BMI category.

Wilcoxon signed-rank tests showed that there were no significant discrepancies in accuracy scores for either the persons with Prader-Willi syndrome or their parents (see Table 4). Fisher's exact test shows that none of the demographic factors were associated with accuracy score. Furthermore, accuracy score was analyzed using multiple regression with BMI, gender, age, IQ, growth hormone therapy status, and genetic subtype as independent variables. For participants with Prader-Willi syndrome, backward selection identified gender, age, and BMI to be significant predictors at a level of 0.1. There also were significant interactions between BMI, age, and gender. To better understand the interactions, we created plots of predicted accuracy versus BMI by gender for younger and older participants separately. Male and female children with Prader-Willi syndrome tended to underestimate their current silhouette as BMI increased in a similar manner (see Table 5 and Figure 4). For the adult group, although female participants' accuracy scores did not change significantly with BMI, male participants tended to underestimate their current BMI as BMI increased (see Table 5 and Figure 5). To examine whether there were any systematic differences between the children and adults with Prader-Willi syndrome, we compared BMI, IQ, gender, age, growth hormone therapy status, and genetic subtype between age groups (< 20 and ≥ 20 years old) and growth hormone status were found to be significantly different between age groups (Table 6). From the preliminary analysis, growth hormone was not associated with the outcome variable accuracy. BMI was included in the interaction analysis as a covariate.

Finally, a one-sample t test was used to test the difference in mean body dissatisfaction score between the adult Prader-Willi syndrome participants and controls from Bulik et al. (2001), stratified by age and gender. Although the participants with Prader-Willi syndrome ($M = 2.07$, $SD = 1.44$) tended to have lower body dissatisfaction than did controls ($M = 2.63$, $SD = 1.3$), the difference was not big enough to reject the null hypothesis at the 5% significance level.

To evaluate whether individuals with Prader-Willi syndrome have the ability to complete the Figure Rating Scale, we analyzed the discrepancy between the child and his or her parent's look scores to determine whether it was associated with the child's IQ. It was not significantly correlated with IQ.

Discussion

These data indicate that responses on the Figure Rating Scale by persons with Prader-Willi syndrome showed significant discrepancies between how they thought they looked and how they wished they looked, not unlike responses of the typical population. Similar discrepancies on the Figure Rating Scale were also found for their parents. In addition, BMI and gender were found to be significant factors on the measure of body dissatisfaction of persons with Prader-Willi syndrome, with females tending to have greater dissatisfaction as BMI scores increased. For parents, however, gender was the only variable with a significant association with body image; parents of males had a greater discrepancy between what they thought their child looked like and what they wished their child looked like.

Further, both the individuals with Prader-Willi syndrome and their parents were fairly accurate on their reports of what they looked like with respect to BMI. When demographic variables were examined, however, there were significant effects of BMI, gender, and age on accuracy.

As BMI increased, both male and female children tended to underestimate their actual silhouette, while adult males were more likely to identify themselves with a smaller silhouette when compared with female adults.

These data are important in that they provide the first evaluation of the how persons with Prader-Willi syndrome describe or perceive their bodies and whether they experience body dissatisfaction. Although adults with Prader-Willi syndrome in our sample tended to have higher body dissatisfaction than did controls, the groups were not significantly different. This should be interpreted with caution due to the small sample size, but it does lend support to the use of the Figure Rating Scale with individuals who have Prader-Willi syndrome. Further support was demonstrated by the comparison between the accuracy score of the participants with Prader-Willi syndrome and their parents' responses. Our analysis also demonstrated that IQ is not a factor in the accuracy of the reporting by persons with Prader-Willi syndrome. In addition, these data indicate that these people are similar to persons without Prader-Willi syndrome in that as their BMI score increased, body dissatisfaction scores increased. In addition, demographic variables in people with Prader-Willi syndrome (i.e., gender, age) were also found to be moderating factors for body image similar to that for the general population.

To evaluate the significance of our results, we have to identify some criteria regarding "poor" body image measurement because there are currently no published standards for what body dissatisfaction score constitutes a serious psychological concern. Bulik et al. (2001) determined that persons who identified themselves with a Number 1 and 2 silhouette typically desired to be heavier, those who chose a 3 were closest to being satisfied. The majority of our participants (91%) chose a 4 or above as the silhouette that represented how they looked. The average body dissatisfaction for our participants who chose a 3 as the silhouette they looked most like was $-.67$, indicating a fairly positive body image.

There is a growing body of literature demonstrating that the use of growth hormone for the treatment of growth hormone deficiency in persons with Prader-Willi syndrome has demonstrated positive effects on stature and muscle mass (e.g., Lindgren et al., 1997) as well as positive psychological effects (e.g., Bertella et al., 2007; Hoybye, Thoren, & Bohm, 2005). Thus, we were interested in whether a person taking growth hormone would show differential effects on their body dissatisfaction scores than would those not taking growth hormone. Our data did not show any difference between the two groups. A variety of uncontrolled variables (e.g., duration of use), however, may have influenced these results. Nevertheless, these results may indicate that despite those on growth hormone therapy feeling better about themselves, their body satisfaction is unaffected by this therapy. Researchers should examine this question further while controlling for variables related to growth hormone, such as length of use.

There are several limitations to this study. First, there were a relatively small number of participants due to the rarity of the disorder, which makes the findings difficult to generalize to the Prader-Willi syndrome population as well as to people with obesity. Despite these concerns, our results did demonstrate a high degree of similarity to other research on this topic.

Second, BMI was not converted into percentiles for the participants under age 18 as is typically done because we wanted to analyze BMI as a continuous variable by age. Future researchers may want to conduct separate data analyses for adults and children with Prader-Willi syndrome to allow for a more accurate comparison with age-matched controls.

Third, these results should also be interpreted with caution because only 77% of the participants had completed intelligence testing, making it difficult to control for cognitive delays. Unfortunately, we did not determine degree of comprehension by the participants with Prader-Willi syndrome to ensure that they understood the Figure Rating Scale. Although this may be of concern due to the mild cognitive delays of this population, the consistency of results when

compared to that of the general population and to the results obtained from the parents of participants with Prader-Willi syndrome makes interpretation of these data less problematic. Future investigators may also want to include an age-matched comparison group of persons with intellectual disabilities due to causes other than Prader-Willi syndrome to determine whether intellectual capacity was a factor in these results. Although normative data were available from Bulik et al. (2001), data were only available for participants 18 and older and were categorized in 10- to 12-year cohorts. Also, the comparison between the participants with Prader-Willi syndrome and the normative data showed a trend, but was not statistically significant. Further investigation on comparison to other groups is needed.

Poor body image or body dissatisfaction is one of the most common factors in poor treatment outcomes for weight loss programs (Carels, Cacciapaglia, Douglass, Rydin, & O'Brien, 2003). Therefore, assessing body dissatisfaction may be an important factor when developing weight loss or maintenance programs for persons with Prader-Willi syndrome. White et al. (2004) showed that parents' views may play a significant role in weight loss efforts as well. Killion, Hughes, Wendt, Pease, and Nicklas (2006) demonstrated that parents felt increasingly less satisfied with their child's body as his or her weight increased, which is consistent with the results in our analysis for the female participants. Little is known about the influence a parent's poor perception of their child's body might play in the success of that child's weight loss or maintenance. However, it stands to reason that, particularly for persons with intellectual and developmental disabilities, a parent's view might play a greater role in their child's body satisfaction and success with interventions than would occur in the typical population. Finally, psychological adjustment, attitude, expectations of success, and proper modeling of dietary practices by parents are associated with successful weight loss in children (White et al. 2004); thus, it may be important for researchers in future studies to include measures of psychological well-being to evaluate how negatively affected individuals with Prader-Willi syndrome are by their weight. For example, the inclusion of measures of depression may be appropriate because disturbances in body image are often correlated with high depression scores (McCabe, Ricciardelli, Sitram, & Mikhail, 2006).

Finally, more research is needed to determine whether it is easier to implement weight management strategies for persons with Prader-Willi syndrome if the individual has an accurate concept of their own body (Foster et al., 1997) or a positive body image.

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Figure 1.

Percentage of adult participants who were obese, overweight, and typical weight and percentage of children who were overweight, at risk of overweight, and healthy, according to the Centers for Disease Control website healthy weight.

Figure 2.

Model of the interaction between body mass index (BMI) and gender and their association with BMI on the body image of persons with Prader-Willi syndrome.

Figure 3.
Mean body dissatisfaction score of adults and children (19 and younger) with Prader-Willi syndrome by weight category.

Figure 4. Regression analysis of degree of accuracy versus body mass index (BMI) by gender for children with Prader-Willi syndrome.

Figure 5. Regression analysis of degree of accuracy versus body mass index (BMI) by gender for adults with Prader-Willi syndrome.

Table 1Demographic Characteristics of Participants ($N = 43$) With Prader-Willi Syndrome

Demographic	<i>n</i> /Mean	%/Range/ <i>SD</i>
Gender (<i>n</i> , %)		
Male	15	35
Female	28	65
Age in years (<i>n</i> , range)	19	10–47
Male	21	11–47
Female	19	10–39
Mean BMI ^a (<i>SD</i>)	31.4	9.0
Male	30.2	7.8
Female	32.4	9.5
BMI classification ^b (<i>n</i> , %)		
Children (19 or younger, <i>n</i> = 22)		
Overweight	11	48
At risk of overweight	5	24
Healthy	6	28
Adult (20 or greater)		
Obese	14	68
Overweight	5	23
Normal weight	2	9
Mean IQ (<i>SD</i> ^c)		
Range	66	13.3
Male (<i>n</i> = 11)	62	10.8
Female (<i>n</i> = 22)	68	14.2
Genetic subtype (<i>n</i> , %)		
Deletion	17	52
Uniparental disomy	14	42
On growth hormone therapy (<i>n</i> , %) ^c	23	70

^aBody mass index.^bBased on Centers for Disease Control criteria.^c $N = 33$ participants from large study of compulsive behavior.

Table 2

Body Dissatisfaction Scores for Persons With Prader-Willi Syndrome (PWS) and Their Parents

Respondent/Category	Median score	Range
Persons with PWS		
Current figure (look)	6.00	2–9
Ideal figure (wish)	4.00	2–6
Body dissatisfaction (look – wish)	2.00	–2–6**
Parents of persons with PWS		
Current figure (look)	6.00	1–9
Ideal figure (wish)	3.00	1–6
Body dissatisfaction (look – wish)	2.00	–3–6***

**
 $p < .01$.

 $p < .001$.

Table 3

Factors Associated With Body Image Dissatisfaction of Persons With Prader-Willi Syndrome

Factor	Slope	95% CI ^a
Interaction between BMI ^b and Gender		
BMI	0.13	0.10, 0.16**
Gender	2.39	0.35, 4.44*
BMI/Gender	-0.06	-0.13, 0
Association between dissatisfaction (parent look – parent wish) Gender	1.55	0.18, 2.93*

^a Confidence interval.^b Body mass index.* $p < .05$.** $p < .01$.

Table 4

Measures of Accuracy

Respondent/Category	Mean	SD
Persons with PWS ^a (<i>n</i> = 43)		
Current figure (look)	6.09	1.56
Accurate figure (BMI ^b)	6.06	2.23
Accuracy (look – BMI)	0.04	1.28
Parents (<i>n</i> = 42)		
Current figure (look)	5.60	1.94
Accurate figure (BMI)	6.01	2.24
Accuracy (look – BMI)	–0.42	1.99

^aPrader-Willi syndrome.

^bBody mass index.

Table 5

Factors Associated With Degree of Accuracy for Persons With Prader-Willi Syndrome (PWS)

Interaction	Slope estimate	95% CI ^a
Interaction between BMI ^b /gender and BMI/age for persons with PWS		
BMI	-0.14	-0.23, -0.05**
Gender	3.82	1.87, 5.77**
Age	-0.17	-0.30, -0.04**
BMI/gender	-0.10	-0.16, -0.04**
BMI/age	0.00	0.00, 0.01*

^a Confidence interval.^b Body mass index.* $p < .05$.** $p < .01$.

Table 6

Factors Associated With Degree of Accuracy

Demographic	<u>Age < 20 (n = 22)</u>		<u>Age ≥ 20 (n = 21)</u>	
	<i>n</i> /Mean	%/ <i>SD</i>	<i>n</i> /Mean	%/ <i>SD</i>
Gender (<i>n</i> ,%)				
Male	7	32	8	38
Mean BMI ^a (<i>SD</i>)	28.0	7.6	35	9.0*
Mean IQ ^b (<i>SD</i>)	63.3	14.6	68.2	11.3
Subtype ^b (<i>n</i> , %)				
Deletion	8	47	9	64
Growth hormone status (<i>n</i> , %) ^b	15	68	8	38*

^aBody mass index.

^b*N* = 33 participants from large study of compulsive behavior.

*
p < .05.