



Who reports it best? A comparison between parent-report, self-report, and the real life social behaviors of adults with Williams syndrome



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ABSTRACT

Given the reliance on self-report in studies of adults with intellectual disabilities, this study examined individual vs. parental reports concerning the social approach behaviors of adults with Williams syndrome (WS) across a hypothetical and a live behavioral setting. Individuals with WS ($N = 30$) were asked whether they would approach strangers in two hypothetical, laboratory tasks (yes/no questionnaire vs. judging facial stimuli of individuals with different emotional expressions). Similarly, their parents also responded to a rating scale of their child's social approach behavior toward strangers displaying various emotions. Then, in a community setting, behavioral coders recorded actual social approaches of individuals with WS toward strangers. Although self-report ratings were consistent across measures, these measures did not correspond to the individuals' actual behaviors during the community observations. Conversely, parental reports did not correspond to their child's self-report measures, but parents did more accurately predict their child's real-life social approach behaviors. Implications are discussed for both research and practice.

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1. Introduction

Due to various methodological issues, the validity and reliability of both parent and self-report questionnaires have recently been questioned. [Finlay and Lyons \(2001\)](#) noted that, for parent reports, researchers usually assume that parents are aware of the individual's behaviors, thoughts, and feelings and are able to accurately report such phenomena. In the emerging area of participatory research, however, researchers have emphasized the importance of obtaining information directly from individuals with disabilities ([Chappell, 2000](#)). But limitations also exist when gathering self-report data from these individuals. Specifically, individuals with intellectual disabilities have difficulty with responding to specific question content, phrasing, and response formats and are prone to inconsistent reporting and acquiescence bias ([Finlay & Lyons, 2001](#); [Heal & Sigelman, 1995](#)). Individuals with intellectual disabilities may also find it difficult to reflect on abstract concepts, including judging their own behaviors or what they would do in hypothetical situations ([Finlay & Lyons](#)).

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Due to these limitations, it remains unclear whether parent or self-report is the most accurate form of measurement related to individuals with intellectual disabilities. While researchers agree that individuals with intellectual disabilities should be given the opportunity to provide their own responses, accommodations are still needed to enhance the accuracy and assess the validity of self-report data. Specifically, [Heal and Sigelman \(1995\)](#) have suggested that multiple formats of self-report data should be collected and compared for response consistency. These self-report data should also be triangulated with reports from other respondents, such as parents, to assess correspondence ([Heal & Sigelman, 1995](#)). [Finlay and Lyons \(2001\)](#) also recommended simplifying the response format from a Likert-type scale to yes/no responses and to provide picture representations with the questions. Finally, in order to determine the most accurate form of measurement, self-report and parent-report data should be supplemented with naturalistic observations ([Schroeder, Cappadocia, Bebko, Pepler, & Weiss, 2014](#)).

In this study, we compared self-report and parent-report data – for both hypothetical, laboratory tasks and for real-life behaviors – of young adults with Williams syndrome. Williams syndrome (WS) is a rare genetic disorder caused by a microdeletion of at least 24 genes on chromosome 7, resulting in borderline to moderate levels of intellectual disability ([Hillier et al., 2003](#); see [Martens, Wilson, & Reutens, 2008](#), for review). These individuals have long been described as having a strong social drive and desire to approach and interact with strangers ([Doyle, Bellugi, Korenberg, & Graham, 2004](#); see [Järvinen, Korenberg, & Bellugi, 2013](#), for review). Statements concerning the social approach behaviors of adults with WS, however, have generally been inferred from parent-report questionnaires and hypothetical self-report tasks ([Fisher, Moskowitz, & Hodapp, 2013](#); [Jones et al., 2000](#); [Riby, Kirk, Hanley, & Riby, 2013](#)).

When results from parent-report are compared to those of self-report, conflicting findings arise as to whether individuals with WS are truly indiscriminate toward approaching strangers. Compared to parents of individuals with other disabilities and individuals without disabilities, parents rate their children with WS as more indiscriminately social (see [Järvinen et al., 2013](#); [Jones et al., 2001](#), for review). But individuals with WS vary in their own reports, with some appearing social and others appearing more socially reserved ([Bellugi, Adolphs, Cassady, & Chiles, 1999](#); [Frigerio et al., 2006](#); [Jones et al., 2000](#); [Martens, Wilson, Dudgeon, & Reutens, 2009](#); [Porter, Coltheart, & Langdon, 2007](#); see [Jones et al., 2001](#), for review).

It is important to note, however, that the self-report data collected from individuals with WS might not be accurate because of the various measurement limitations associated with assessing individuals with intellectual disabilities. For example, individuals with WS are often asked to rate their own social approach behavior in hypothetical situations, such as through viewing photographs of unknown adults and indicating whether they would like to approach and interact with the person (see [Jones et al., 2001](#), for review). These responses should be interpreted with caution, as individuals with intellectual disabilities have a difficult time responding to questions about abstract concepts, such as hypothetical situations ([Heal & Sigelman, 1995](#)). Additionally, participants with WS are typically asked to rate their desire to approach on a 4 to 5-point Likert-scale; yet, individuals with intellectual disabilities have a difficult time providing answers to Likert-type scales ([Finlay & Lyons, 2001](#)). Finally, participants are only administered one assessment of approachability, forfeiting the possibility of checking for consistency across measures ([Heal & Sigelman](#)).

A final limitation is that only one study has directly compared parent-report to self-report and no studies have compared these reports to naturalistic observations of the social behaviors of individuals with WS. When [Järvinen-Pasley et al. \(2010\)](#) directly compared parent-report to self-report within the same sample, they found no correspondence between responses. Thus, it remains unclear whether parents or individuals with WS are more accurate at reporting the social behaviors of adults with WS.

The current study had three aims. First, to assess the consistency of reporting among adults with WS, we compared two forms of self-report data, concerning social approach behavior. To account for a possible effect of social desirability (e.g., participants providing the response they thought we were looking for), one task was administered through a computer program with no researcher interaction, while the other was administered verbally with a research assistant. Additionally, to further comply with the recommendations for self-report assessment with individuals with intellectual disabilities, we simplified the response format from a Likert-scale to yes/no ([Finlay & Lyons, 2001](#)). Second, we assessed the correspondence between parent-report and self-report by comparing both forms of self-report measurements to the parents' responses to similar questions concerning the social approach behaviors of adults with WS. Finally, to determine the most accurate reporter of social approach behaviors, we compared parent-report and self-report to a naturalistic observation of the social behaviors of the sample of adults with WS.

2. Method

2.1. Participants

Participants consisted of 30 young adults with genetically confirmed WS (15 female, 15 male) and their parents (26 mothers, 4 fathers). All participants were White. Individuals with WS were an average of 26.43 ($SD = 8.56$) years of age (range 16–50). Participants with WS were administered the Kaufman Brief Intelligence Test, 2nd edition (KBIT-2; [Kaufman & Kaufman, 2004](#)), which yields verbal, non-verbal, and full scale IQ scores. Average full scale IQ was 70.43 ($SD = 13.73$); consistent with the WS cognitive profile ([Searcy, Lincoln, Rose, Klima, Bavar, & Korenberg, 2004](#)), the verbal standard score averaged 77.50 ($SD = 11.51$) and the non-verbal standard score averaged 69.80 ($SD = 16.90$). Most individuals with WS lived at home with their parents (90%), while the remaining participants lived independently with support (10%).

The mean age of responding mothers was 54.69 ($SD = 9.62$) and the mean age of responding fathers was 55.67 ($SD = 7.09$). The majority of mothers had earned either a 2- or 4-year degree (34.6%) and 26.9% had earned a graduate or professional degree. All of the fathers had earned a 4-year degree. Mothers worked an average of 20.72 ($SD = 19.75$) hours outside the home, while fathers worked an average of 31.67 ($SD = 28.43$) hours outside the home. About half ($M = 53.3\%$) of the parent respondents were still married to the participant's other parent.

2.2. Measures

2.2.1. Demographics Questionnaire

Demographics Questionnaire collected basic information about the individual with WS, including age, living situation, parent's occupation, and estimated family income.

2.2.2. Kaufman Brief Intelligence Test, 2nd edition

Kaufman Brief Intelligence Test, 2nd edition (KBIT-2; Kaufman & Kaufman, 2004) is a psychometric measure for individuals aged 4–90 years. The KBIT-2 includes a composite IQ as well as standard scores for verbal and non-verbal knowledge. The KBIT-2 has been used with persons with Williams syndrome in previous studies (e.g., Dykens, Rosner, & Ly, 2001; Mervis, Kistler, John, & Morris, 2012).

2.2.3. Self-Report Approachability Scale

Self-Report Approachability Scale (Approachability Scale) was created for this study. It consisted of eight self-report questions related to participant's social approach behaviors. A research assistant asked the participant, "I am going to ask you whether or not you would like to go up and talk to different types of people you do not know. There are no right or wrong answers. Please answer each question 'yes' or 'no'. If you can't decide, think about what you would probably do in each situation. Would you like to go up and talk to a (woman/man) (you do not know/who looks happy/sad/angry)?" "Yes" responses were given a score of "1" and "no" responses were given a score of "0". While this dichotomization of the response options limits response variability and the ability to compare responses to parent-report, we felt it was prudent to attempt to obtain the most reliable response from the participant. A total score of how many people the participant would approach was then calculated, ranging from 0 (the participant would not approach any of the strangers) to 8 (the participant would approach all of the strangers).

2.2.4. Self-Report Faces Task

Self-Report Faces Task (Faces Task) was similar to previous assessments of self-report approachability behaviors used with individuals with WS (e.g., Frigerio et al., 2006). Specifically, participants viewed 144 color photographs of unfamiliar faces depicting 36 young adults displaying happy, sad, angry, and neutral expressions, obtained from a standardized Radboud Faces Database set (Langner, Dotsch, Bijlstra, Wigboldus, Hawk, & van Knippenberg, 2010). Photographs were presented on a computer screen in random order using E-prime (v.2.0, PST Inc., Pittsburgh, PA). Using this format, participants were not required to interact with the researcher, thus reducing the potential of social desirability response biases. At the beginning of the task, participants viewed instructions on the screen asking them to indicate whether they would like to walk up and talk to the person by pressing either a "yes" or "no" button (instructions were also read aloud to all participants). Responses were given a score of "1" if the participant indicated a desire to approach and "0" if the participant did not indicate a desire to approach. Percentages of positive responses were then calculated for each emotion, ranging from 0% (the participant would not approach any of the strangers displaying the specific emotion) to 100% (the participant would approach all of the strangers displaying the specific emotion).

2.2.5. Parent Report Approachability Scale

Parent Report Approachability Scale (Parent-Report) was created to mirror the Self-Report Approachability Scale. Specifically, it was designed to assess parent's perceptions of the social approach behavior of individuals with WS. Through a series of 8 questions, parents were asked to rate on a scale of 1 (*not very likely*) to 5 (*very likely*), "How likely is your child to approach a (male/female) who looks (unfamiliar/happy/sad/angry)?" A total score of how many people the child would approach (rating of 4 or higher) was then calculated, ranging from 0 (the child was unlikely to approach any of the described strangers) to 8 (the child would likely approach all of the described strangers).

2.3. Procedure

This project was part of a larger research study being conducted with adults with WS attending a weeklong residential summer camp program. Parents of participants were asked if they would like their child to participate in research while attending the camp. Interested parents signed a consent form and the individuals with WS signed an assent form. The study was conducted with approval from the University's Institutional Review Board.

Prior to arrival at camp, parents were emailed the link to the demographics questionnaire and Parent Report Approachability Scale, which were included as part of a larger online survey. Upon arrival at camp, participants with WS were scheduled to complete various research tasks. As part of this scheduling, they first met with a research assistant in a

private room to complete the KBIT-2. Next, they completed the Faces Task and then immediately met with a research assistant to complete the Approachability Scale. All participants completed the tasks in the same order. The decision was made to administer the Faces Task prior to the Approachability Scale to account for the disagreement in the literature concerning whether individuals with WS have difficulty labeling emotions (Martens et al., 2008). Specifically, the Faces Task was used as a way to prime participants to respond to questions about emotions without the aid of a visual representation of each emotion.

At least two days after administration of the laboratory tasks, participants' behaviors were observed and coded in a community setting while they enjoyed dinner and dancing at a local restaurant. During this outing, five behavioral coders were employed to observe and record the participants' interactions. Each coder was assigned to observe and collect data on a group of 6 participants using partial interval time sampling. Specifically, within each group, one participant was observed by the coder every 30 s. The 30-s intervals were signaled through a MotivAider®. This device (much like a pager) was clipped on the pants pocket of each observer and was preset to vibrate every 30 s. Upon the vibration signal, the coder briefly looked down at the data collection sheet and checked each type of individual with whom the participant interacted during the interval (e.g., female/male camper, female/male counselor, female/male stranger, no interaction). Following documentation, the observer looked up and began observing the next participant for 30 s. This process continued for each participant, every 30 s for 1.5 h, producing a total of 24 observation intervals for each participant.

Inter-observer reliability data, while crucial to observational data, could not be collected for logistical reasons of the study (e.g., the number of personnel required would have doubled the size of the camp personnel and raised speculation among the participants). Because of this, we took many steps to ensure the behavioral coders were well trained prior to data collection and had ways to verify behaviors during the observation session. Specifically, all behavioral coders were trained in data collection procedures during a 1-h training, prior to the start of the research study. During this training, coders were provided with the definition of a social interaction, "any instance of a camper talking to, making physical contact with, or looking at someone talking to them for at least 3 s. They must be no more than 5 feet from the person with whom they are interacting". Examples included "standing in a group and talking to one person or giving someone a high five without looking at them, dancing with someone, and sitting at a table talking to someone". Non-social examples included "looking across the room at someone, sitting alone at a table, or staring into an empty space." They were also given the definition of a stranger, defined as, "anyone not associated with the camp or the research study. This includes anyone not wearing a camp nametag, including other patrons at the restaurant, special guests, personnel with the media, wait staff, band members on stage, and the DJ. This did not include counselors, campers, research assistants, or anyone else wearing a camp nametag". Coders were asked to read the definitions and to ask for clarification. The first author then asked coders to judge whether certain descriptions fit within the definition of social interaction and stranger (e.g., "what if the camper walks up to the waiter and asks for a coke?").

Finally, at the end of training, coders were provided with pictures of each camper, the written definitions of social interactions and strangers, a clipboard, and data collection sheets. They were instructed to arrive at the restaurant approximately 30 min before the planned arrival of the participants. This allowed the coders to find an optimal location for observations. During the 1.5-h observation session, the first author was available to answer any questions about the target definitions or to help locate specific participants. After data collection, the first author met with the coders to discuss the observations and clarify any questions about specific observations that might have arisen. The coders had very few questions, as they felt the definitions were straightforward and they were easily able to judge whether participants were interacting and with which type of person.

2.4. Data analysis

Due to the small sample size for certain analyses, dichotomous and non-continuous variables of interest, and large standard deviations, it was apparent that non-parametric descriptive and inferential statistics should be used instead of the more conventional means, correlations, and *t*-tests (Nanna & Sawilowsky, 1998; Siegel, 1956).

2.4.1. Preliminary analyses

Wilcoxon signed ranks test were used to determine whether responses on the Self-Report Approachability Scale, Self-Report Faces Task, and Parent Report Approachability Scale differed by gender of the stranger and to compare how gender of the stranger and gender of the participants related to approach behavior. Friedman's chi-square was used to determine if responses differed by gender of the individual with WS. The remaining analyses were conducted with participant's average response across genders because there were minimal differences. Kruskal–Wallis analyses were used to examine the differences in the percent of "yes" responses to happy, sad, and angry faces on the Self-Report Faces Task, compare responses to different strangers' emotions on the Parent Report Approachability Scale, and examine whether participants displayed differences in their approach behaviors toward female campers, counselors, and strangers and male campers, counselors, and strangers.

2.4.2. Consistency in reporting across measures

To examine correspondence between the Approachability Scale and Faces Task, we first selected those participants who answered, "Yes, I would approach" to at least one gender for each emotion on the Approachability Scale and then calculated the mean "yes" responses on the Faces task. Next, we conducted the same analyses, but with those who said "no" to each emotion on the Approachability Scale.

Table 1

Analyses conducted for the various comparisons.

| Measure comparison | Score conversion | Statistic |
|--|--|---|
| Approachability Scale vs. Faces Task | Select only “Yes” on Approachability Scale compared to % yes on Faces Task | — |
| Approachability Scale vs. Faces Task | Select only “No” on Approachability Scale compared to % yes on Faces Task | — |
| Parent-Report vs. Approachability Scale | Parent measure dichotomized to yes/no | Kappas |
| Parent-Report vs. Approachability Scale | Total scores (0–8) for both measures | Spearman's rho correlation |
| Parent-Report vs. Faces Task | Convert percent on Faces Task into ranks | Spearman's rho correlation |
| Approachability Scale vs. Community Observations | Total number of strangers approached | Mann–Whitney <i>U</i> Spearman's Rho correlation |
| Faces Task vs. Community Observation | Calculated percent of observed intervals out of 24) in which participant approached stranger | Spearman's rho correlation with percent “yes” to neutral expression on Faces task |
| Parent-Report vs. Community Observation | Total score on parent report and total number of strangers approached | Mann–Whitney <i>U</i> Spearman's Rho correlation |

To compare parent report to participant responses on the Approachability Task, parent responses were scored as 1, “Yes, the individual with WS would approach” if the parent rated the item as 4 or higher, and 0, “No, the individual would not approach” if the parent rated the item less than 4. If the participant indicated a desire to approach at least one gender within an emotion, the response was scored as 1, “Yes, I would approach”. If the participant did not want to approach either gender within an emotion, the response was scored as 0. We then calculated Kappas to compare the parent and participant yes/no responses. Bivariate correlations were used to explore the relationship between total scores on both measures. To compare parent report to the Faces task, we converted the percentage scores on the Faces task into ranks (1 [0–20%], 2 [21–40%], 3 [41–60%], 4 [61–80%], and 5 [81–100%]) to correspond with the parent report rankings of 1 (not very likely to approach) to 5 (very likely) and then conducted Spearman's rho correlations.

2.4.3. Correspondence with real life behavior

For the Approachability Task, a Mann–Whitney *U* test was used to compare responses to the questions “Would you like to go up and talk to a woman/man you do not know” to the total number of strangers approached during the community observation. Additionally, a bivariate Spearman's rho correlation was conducted to compare the total score on the Approachability Task to the total number of strangers approached. For the Faces Task, we first determined the percent of observed intervals (out of 24) in which the participant approached a stranger and then used Spearman's rho to correlate those scores with the average percent of times in which the participant said “Yes” to approaching a picture of a stranger expressing no emotion (neutral expression). Finally, similar to the Approachability Task, a Mann–Whitney *U* test was used to compare the dichotomized parent responses to the questions “How likely is your child to approach a male/female who looks unfamiliar” to the total number of strangers approached during the community observation. We also used Spearman's rho to correlate the parent's total score on the Parent Report Task to the total number of strangers the participant approached during the community observation (see Table 1 for more details on score conversions and analyses).

3. Results

3.1. Preliminary findings

3.1.1. Self-Report Approachability Scale

Participant responses varied by emotion of the stranger. Specifically, 90% of participants indicated they would approach a happy stranger, no participant (0%) indicated a desire to approach an angry stranger, and very few participants (13%) indicated that they would like to approach a stranger without emotion. Responses were mixed concerning approaching a sad stranger (47% said yes). Friedman's test revealed participants' desire to approach a stranger differed by emotion ($\chi^2[30] = 42.00$, $p < .001$). Post hoc analysis with Wilcoxon signed-rank tests revealed participants were significantly more likely to indicate a desire to approach a happy stranger, compared to a sad stranger ($z = -3.56$, $p < .01$) or an angry stranger ($z = -5.14$, $p < .01$). Also, participants were significantly more likely to indicate a desire to approach a sad stranger than an angry stranger ($z = 3.74$, $p < .01$). Responses did not differ based on the gender of the stranger (see Table 2), nor by gender of the participant.

3.1.2. Self-Report Faces Task

When viewing neutral facial expressions, participants with WS rarely indicated they would like to approach and talk to the stranger (average 17% yes [$SD = 16\%$], range 0–63%). Friedman's test revealed participants' desire to approach a stranger differed by emotion ($\chi^2[30] = 48.81$, $p < .001$). Post hoc analysis with Wilcoxon signed-rank tests revealed participants were significantly more likely to indicate a desire to approach a happy stranger (88% yes), compared to a sad stranger (11%; $z = -4.79$, $p < .01$) or an angry stranger (7%; $z = -4.79$, $p < .01$). Similar to self-report without stimuli, responses did not differ based on gender of the stranger or gender of the individual with WS.

Table 2
Differences in parent reported approach behavior and self-reported approach behavior based on the gender and emotion of the stranger.

| Gender of stranger | Male | Female | <i>z</i> |
|--|-------------|-------------|----------|
| Self-Report Approachability Scale | | | |
| Happy | .87 (.35) | .93 (.25) | –1.44 |
| Sad | .47 (.51) | .47 (.51) | .00 |
| Angry | .00 (.00) | .00 (.00) | .00 |
| Unknown | .07 (.25) | .13 (.35) | –1.41 |
| Parent Report Approachability Scale | | | |
| Happy | 3.19 (1.39) | 3.63 (1.42) | –2.97** |
| Sad | 2.96 (1.22) | 3.70 (1.30) | –3.22** |
| Angry | 1.30 (.67) | 1.67 (1.00) | –2.23* |
| Unknown | 2.52 (1.28) | 2.74 (1.40) | –1.27 |

* $p < .05$.

** $p < .01$.

3.1.3. Parent Report Approachability Scale

Parents rated adults with WS as only somewhat likely to approach an unfamiliar person ($M = 2.63$, $SD = 1.25$, range 1.00 to 4.50). Again, Friedman's test revealed the emotion of the stranger was significantly related to the approachability rating ($\chi^2[27] = 38.67$, $p < .001$), with parents indicating their child was more likely to approach a stranger who looked happy ($M = 3.41$ [$SD = 1.37$]) or sad ($M = 3.33$ [$SD = 1.17$]) compared to angry ($M = 1.48$ [$SD = .75$]); z 's = -4.21 and -4.33 , respectively, both p 's $< .01$). The gender of the stranger was also important. Parents indicated their child was more likely to approach females displaying all emotions compared to males (see Table 2). Responses did not differ by gender of the individual with WS.

3.1.4. Community observations

When observed in a community setting, individuals with WS did not often engage with strangers; participants spent an average of 42% of time not interacting with anyone. There was a statistically significant difference in number of females and males approached, when comparing whether they were campers, counselors, or strangers (χ^2 's [2] = 6.91 and 31.14, $p < .05$), respectively. Post hoc analysis with Wilcoxon signed-rank tests revealed the participants engaged with more female participants ($M = 4.17$ [$SD = 4.04$]) and more female counselors ($M = 5.28$ [$SD = 4.12$]; $z = -3.31$, $p < .01$) than female strangers ($M = 2.14$ [$SD = 1.55$]), $z = -2.20$, $p < .05$. Participants more often engaged with male campers (5.34 [4.88]), than with male counselors ($M = 1.03$ [$SD = 1.30$]) $z = -4.21$, $p < .01$) or male strangers ($M = 1.10$ [$SD = 1.54$]), $z = -4.20$, $p < .01$. Finally, a Wilcoxon signed-rank test revealed that participants were more often engaged with female strangers ($M = 2.13$ [$SD = 1.55$]) compared to male strangers ($M = 1.10$ [$SD = 1.54$]), $z = -2.56$, $p < .01$. The gender of the participant was not related to the gender of person with which they were likely to engage.

3.2. Consistency in reporting across measures

3.2.1. Approachability Scale vs. Faces Task

There was good correspondence across both measures and participants indicating a desire to approach a stranger with a happy emotion. The 28 participants who said "yes" to approaching a happy person on the Approachability Scale, said "yes" to an average of 87% of the happy pictures, providing evidence of correspondence between the two measures. On the other hand, the 14 participants who said, "yes" to approaching a sad person on the Approachability Scale said, "yes" to an average of only 11% of sad pictures on the Faces Task. The four participants who indicated they would approach someone they did not know on the Approachability Scale, only said "yes" to an average of 34% of the neutral pictures on the Faces Task, indicating a lack of correspondence between measures (see Table 3).

The two participants who said "no" to a happy stranger on the Approachability Scale responded "yes" to 99% of the pictures on the Faces Task. Thus, there is no correspondence between those who said "no" on the Approachability Scale and their performance on the Faces task for happy strangers.

There was a high correspondence between indicating avoidance of negative emotional expressions on both measures. Those participants, who responded "no" to sad and angry people on the Approachability Scale, responded similarly on the Faces task. The 16 participants who said "no" to approaching a sad person on the Approachability Scale indicated, "no" to 88% of the sad pictures on the Faces Task. No participants said they would approach an angry stranger on the Approachability Task and they similarly indicated "no" to angry Pictures 93% of the time on the Faces Task. Finally, the 16 participants who said "no" to an unknown individual on the Approachability Scale indicated they would not approach a neutral face, saying "no" 86% of the time on the Faces task (Table 3).

3.3. Correspondence between parent report and self report

Because there was no systematic difference in responses on the Approachability Task and the Faces Task, the remaining comparisons were conducted only with the Approachability Task.

Table 3

Top: Mean “yes” responses to pictures of emotions on the Faces Task for those participants who answered “Yes” to specific emotions on the Self-Report Approachability Scale. Bottom: Mean “yes” responses to pictures of emotions on the Faces Task for those participants who answered “no” to specific emotions on the Self-Report Approachability Scale.

| | N | Min | Max | Mean |
|------------|----|-----|------|-----------|
| Yes | | | | |
| Happy | 28 | .42 | 1.00 | .87 (.16) |
| Sad | 14 | .00 | .60 | .11 (.18) |
| Angry | 0 | | | |
| Unknown | 4 | .06 | .63 | .34 (.24) |
| No | | | | |
| Happy | 2 | .97 | 1.00 | .99 (.02) |
| Sad | 16 | .00 | .67 | .12 (.19) |
| Angry | 30 | .00 | .49 | .07 (.11) |
| Unknown | 26 | .00 | .48 | .14 (.14) |

Table 4

Top: Total score on the Approachability Scale compared to the number of strangers the participant approached during the community observation. Bottom: Total score on the Parent Report Scale compared to the number of strangers the participant approached during the community observation.

| Total score | N | Mean strangers approached |
|------------------------------|----|---------------------------|
| Approachability Scale | | |
| 0–1 | 4 | 2.75 (.96) |
| 2–4 | 21 | 3.38 (2.40) |
| 5–6 | 4 | 3.00 (4.24) |
| Parent Report Scale | | |
| 0–1 | 3 | 1.67 (2.08) |
| 2–4 | 9 | 2.56 (1.42) |
| 5–8 | 14 | 4.21 (2.89) |

3.3.1. Parent report vs. Approachability Scale

There was no agreement between the parent and the individual with WS on the reported social approach behaviors (Kappas ranged from $-.01$ to $.33$). Regarding total scores, Wilcoxon signed ranks test revealed that parent’s mean score was significantly higher (4.41 [$SD = (2.43)$], range of 0–8) than the mean score for individuals with WS (2.93 [$SD = 1.60$]; range 0–6), $z = -2.59$, $p < .01$. Finally, parent and self-report scores did not correlate ($\rho = .23$, ns), indicating a lack of correspondence between parent and self-report social approach behaviors using the Approachability Scales.

3.4. Correspondence with real life behavior

3.4.1. Self-Report Approachability Scale

There were no differences between those who said they would approach a stranger vs. those who would not approach a stranger and the number of strangers actually approached during the community observation. Specifically, the four participants who said “yes” to approaching a stranger approached an average of 3.0 ($SD = 4.24$) strangers and the 25 who reported “no” approached an average of 3.28 (2.27) strangers, Mann–Whitney $U = 37.50$, ns . Additionally, the total score on the Approachability Scale did not correlate with the total number of strangers approached during the community observation ($\rho [30] = .01$, ns).

3.4.2. Parent Report Approachability Scale

There were no differences between those parents who said their child would approach a stranger vs. those who said their child would not approach a stranger and the number of strangers actually approached during the community observation. Specifically, of the 9 participants whose parent said “yes” they would approach a stranger, those participants approached an average of 4.67 ($SD = 4.20$) strangers and the 17 whose parent reported “no” they would not approach an average, approached an average of 2.33 (2.52) strangers, Mann–Whitney $U = 50.50$, ns . On the other hand, the parent total score on the Parent Report Scale significantly correlated with the total number of strangers the participants approached during the community observation ($\rho [27] = .45$, $p < .05$). To further explore this correlation, we specifically examined each participant’s total scores on the Parent-Report Scale and Approachability Scale and compared them to the number of strangers actually approached (see Table 4). The more strangers the parents reported, the more strangers were actually approached. Comparatively, those participants who said they would not approach strangers approached an average of 2 strangers; those who said they would approach the most strangers actually did not approach and engage any stranger during the community outing.

4. Discussion

In this study, we tackle head on the question of reporter accuracy, the most useful measurement for assessing self-report, and the correspondence between reporting and real life behavior. Assessing the social approach behavior of adults with WS, we first addressed self-report measurement limitations by creating two self-report instruments and examining whether the responses from participants with WS varied based on type of assessment. Next, we explored whether participant responses corresponded with parent responses. Finally, moving beyond parent and self-report, this study was the first to compare these reports to the actual real life behaviors of adults with WS. This study has three main findings.

The first major finding of this study related to the consistency of self-report across two types of measurement. Having first created two assessments, both of which used a simplified yes/no response format, we asked adults with WS to rate the desire to approach and engage strangers displaying various emotional expressions (Finlay & Lyons, 2001). Despite differences in stimulus presentation, participants' responses to emotions on the Faces Task were similar to their responses on the Approachability Scale. Overall, participants indicated they would like to approach a stranger who looked happy and would not approach a person they did not know and a stranger who looked angry. Responses were less consistent related to strangers who were sad. The second major finding of this study related to the lack of correspondence between self-report and parent report. Similar to previous research, parents (compared to adults with WS) more often rated individuals with WS as likely to approach and engage strangers (Järvinen-Pasley et al., 2010). The high social approach ratings could explain why parents are so concerned about whether these social behaviors might lead to exploitation (Fisher et al., 2013). Specifically, compared to their parents, the adults with WS were far less likely to report a desire to approach and engage strangers. The difference in responses may indicate that adults with WS lack the ability to inhibit the desire to engage strangers, even if they have been taught and are able to restate the "rule" that you do not talk to strangers.

The third major finding was that parents are more accurate. Specifically, the number of strangers approached by individuals with WS did not differ based on their response to the Approachability Scale. In fact, the two participants who said they would not approach any stranger on the Approachability Scale both approached two strangers during the observation; alternatively, the two participants with the highest total score on the Approachability Scale (said yes to approaching six strangers) did not approach and engage any strangers during the community observation. Regarding parent report, while the number of strangers approached by individuals with WS did not differ based on whether parents said they would approach or not, parents' high social approach scores were significantly related to the actual number of strangers approached by participants with WS during the community observation. The findings are counter-intuitive to the intellectual disability literature, which generally indicates that individuals with intellectual disabilities should be the main informants related to their own behaviors (Chappell, 2000). As such, our results point to the importance of including parent report in assessments of adults with WS, especially when examining social behaviors.

The consistency in responses between the self-report measures has important implications for both research and practice. First, while previous research has relied on more involved measures of social approach behavior (similar to the Faces task; see Jones et al., 2001 for review), these findings indicate that self-report questionnaires without visual stimuli result in similar responses from adults with WS. A simple paper and pencil questionnaire (like the Approachability Scale) is a less time consuming and more readily accessible measure for researchers or practitioners who wish to gather information about social behaviors directly from individuals with WS. Second, the consistency in the relatively low desire to approach and engage a stranger (13% said yes on the Approachability Scale vs. mean of 16% 'yes' response to neutral pictures on Faces Task) indicates that adults with WS seem to have internalized the message that they should not talk to strangers. This finding also indicates, however, that while participants seem to know what to say, they might not always act appropriately in real situations.

These findings also highlight the importance of acknowledging those parents who report hypersocial behaviors of adults with WS. As parents seem to be more aware of the social behavior of adults with WS than are the individuals themselves, we should work with parents to develop effective ways to teach social safety skills to adults with WS. Additionally, these findings highlight the importance of comparing questionnaire measures to behavioral observations that occur in real world settings. Such observations have served as a way to supplement and validate parent report. For example, if researchers and practitioners rely on a pre-post test assessment of skill acquisition given just to the adult with WS, then it is possible they will not accurately measure real life behaviors. Therefore, once trainings to teach social safety skills are developed and implemented, it will be important to continue to collect data from the parent and through behavioral observations (Fisher et al., 2013).

While this study expanded the research related to the social behaviors of adults with WS by comparing parent and self-report to real life behaviors, certain limitations must be acknowledged. First, different rating scales were used for the parent-report and self-report measures. Because individuals with intellectual disability have difficulty responding to Likert scales (Finlay & Lyons, 2001), we chose to simplify the scale to a yes/no response for the adults with WS. Using two different scales for parent- and self-report led to the need to dichotomize the parent-report scale for many of the analyses, which decreases variability and thus, sensitivity of the measures. Second, the individuals with WS were a part of an overnight camp. In order to attend this camp, participants were required to have few behavior problems, to be able to stay away from home, and to be able to independently complete many adaptive living skills, such as dressing and toileting. As such, these participants might not be a representative sample of adults with WS. Finally, one participant was not available for the community observation and some parents failed to answer all questions on the parent-report questionnaire.

Despite these limitations, this study provides important and novel insight into the most accurate informant of the social behavior of adults with WS. Furthermore, we were able to observe and quantify the social behaviors in a natural, real life situation. We found that while adults with WS are consistent in reporting their behaviors across measures, their responses do not correspond to their real life behaviors. On the other hand, while parent's responses do not correspond with participant responses, parent's responses are consistent with observations in the natural setting. Taken together, these results indicate that even though individuals with WS are somewhat consistent at reporting their own behaviors toward strangers, these responses inaccurately represent the way they behave in real life. Parents, on the other hand, are able to accurately report the way adults with WS would most likely act toward strangers in real life situations.

Conflict of interest

None declared.

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References

- Bellugi, U., Adolphs, R., Cassady, C., & Chiles, M. (1999). Towards the neural basis for hypersociability in a genetic syndrome. *Neuroreport*, *10*, 1653–1657.
- Chappell, A. L. (2000). Emergence of participatory methodology in learning difficulties researcher: Understanding the context. *British Journal of Learning Disabilities*, *28*, 38–43.
- Doyle, T. F., Bellugi, U., Korenberg, J. R., & Graham, J. (2004). "Everybody in the world is my friend": Hypersociability in young children with Williams syndrome. *American Journal of Medical Genetics*, *124A*, 263–273.
- Dykens, E. M., Rosner, B. A., & Ly, T. M. (2001). Drawings by individuals with Williams syndrome: Are people different from shapes? *American Journal on Mental Retardation*, *106*, 94–107.
- Finlay, W. M. L., & Lyons, E. (2001). Methodological issues in interviewing and using self-report questionnaires with people with mental retardation. *Psychological Assessment*, *13*, 319–335.
- Fisher, M. H., Moskowitz, A., & Hodapp, R. M. (2013). Differences in social vulnerability among individuals with autism spectrum disorder, Williams syndrome, and Down syndrome. *Research in Autism Spectrum Disorders*, *7*, 931–937.
- Frigerio, E., Burt, D. M., Gagliardi, C., Cioffi, G., Martelli, S., Perrett, D. I., et al. (2006). Is everybody always my friend? Perception of approachability in Williams syndrome. *Neuropsychologia*, *44*, 254–259.
- Heal, L. W., & Sigelman, C. K. (1995). Response biases in interviews of individuals with limited mental ability. *Journal of Intellectual Disability Research*, *39*, 331–340.
- Hillier, L. W., Fulton, R. S., Fulton, L. A., Graves, T. A., Pepin, K. H., Wagner-McPherson, C., et al. (2003). The DNA sequence of chromosome 7. *Nature*, *424*, 157–164.
- Järvinen-Pasley, A., Adolphs, R., Yam, A., Hill, K. J., Grichanik, M., Reilly, J., et al. (2010). Affiliative behavior in Williams syndrome: Social perception and real-life social behavior. *Neuropsychologia*, *48*, 2110–2119.
- Järvinen, A., Korenberg, J. R., & Bellugi, U. (2013). The social phenotype of Williams syndrome. *Current Opinion in Neurobiology*, *23*, 1–9.
- Jones, W., Bellugi, U., Lai, Z., Chiles, M., Reilly, J., Lincoln, A., et al. (2000). Hypersociability in Williams syndrome. *Journal of Cognitive Neuroscience*, *12*(Suppl. 1), 30–46.
- Jones, W., Bellugi, U., Lai, Z., Chiles, M., Reilly, J., Lincoln, A., et al. (2001). Hypersociability: The social and affective phenotype of Williams syndrome. In U. M. Bellugi, S. George, & A. M. Galaburda (Eds.), *Journey from cognition to brain to gene*. Cambridge, MA: MIT Press.
- Kaufman, A., & Kaufman, N. (2004). *Kaufman brief intelligence test* (2nd ed.). Circle Pines, MN: American Guidance Service.
- Langner, O., Dotsch, R., Bijlstra, G., Wigboldus, D., Hawk, S., & van Knippenberg, A. (2010). Presentation and validation of the Radboud Faces Database. *Cognition and Emotion*, *24*(8), 1377–1388.
- Martens, M. A., Wilson, S. J., & Reutens, D. C. (2008). Research review: Williams syndrome: A critical review of the cognitive, behavioral, and neuroanatomical phenotype. *Journal of Child Psychology and Psychiatry*, *49*(6), 576–608.
- Martens, M. A., Wilson, S. J., Dudgeon, P., & Reutens, D. C. (2009). Approachability and the amygdala: Insights from Williams syndrome. *Neuropsychologia*, *47*, 2446–2453.
- Mervis, C. B., Kistler, D. J., John, A. E., & Morris, C. A. (2012). Longitudinal assessment of intellectual abilities of children with Williams syndrome: Multilevel modeling of performance on the Kaufman Brief Intelligence Test-Second Edition. *American Journal on Intellectual and Developmental Disabilities*, *117*, 134–155.
- Nanna, M. J., & Sawilowsky, S. S. (1998). Analysis of Likert scale data in disability and medical rehabilitation research. *Psychological Methods*, *3*, 55–67.
- Porter, M. A., Coltheart, M., & Langdon, R. (2007). The neuropsychological basis of hypersociability in Williams and Down syndrome. *Neuropsychologia*, *45*(12), 2839–2849.
- Riby, D. M., Kirk, H., Hanley, M., & Riby, L. M. (2013). Stranger danger awareness in Williams syndrome. *Journal of Intellectual Disability Research*. <http://dx.doi.org/10.1111/jir.12055>
- Schroeder, J. H., Cappadocia, M. C., Bebko, J. M., Pepler, D. J., & Weiss, J. A. (2014). Shedding light on a pervasive problem: A review of research on bullying experiences among children with autism spectrum disorders. *Journal of Autism and Other Developmental Disorders*. <http://dx.doi.org/10.1007/s10803-013-2011-8>
- Searcy, Y. M., Lincoln, A. J., Rose, F. E., Klima, E. S., Bavar, N., & Korenberg, J. R. (2004). The relationship between age and IQ in adults with Williams syndrome. *American Journal on Mental Retardation*, *109*, 231–236.
- Siegel, S. (1956). *Nonparametric statistics for the behavioral sciences*. New York: McGraw-Hill Book Company Inc.