Published in final edited form as:

Am J Intellect Dev Disabil. 2017 March ; 122(2): 156–172. doi:10.1352/1944-7558-122.2.156.

Behavioral Profiles of 6 – 14 Year-Old Children with Williams Syndrome from Spain and the United States: Cross-Cultural Similarities and Differences

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Abstract

To identify similarities and differences in the behavioral profile of children with Williams syndrome from Spain and the United States, we asked parents of 6 – 14-year-olds from Spain (n=53) and the United States (n=145) to complete the Child Behavior Checklist 6–18. The distribution of raw scores was significantly higher for the Spanish sample than the American sample for half of both the empirically-based and DSM-oriented scales. In contrast, analyses based on country-specific T-scores indicated that the distribution for the Spanish sample was significantly higher than for the American sample only on the Social Problems scale. No gender differences were found. Genetic and cultural influences on children's behavior and cultural influences on parental ratings of behavior are discussed.

Keywords

Williams syndrome; Williams-Beuren syndrome; intellectual disability; behavior; CBCL; cross-cultural

Introduction

Williams syndrome (WS) is a neurodevelopmental disorder caused by a heterozygous deletion of 1.55 – 1.83 megabases (26 – 28 genes) on chromosome 7q11.23 (Pérez-Jurado, 2003; Bayes, Magano, Rivera, Flores, & Pérez-Jurado, 2003). The neurocognitive profile of individuals with WS is characterized by mild to moderate intellectual disability with a specific weakness in visuospatial construction and relatively strong concrete vocabulary

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skills (Mervis & John, 2010). A distinctive behavioral profile also has been described, including high levels of social approach/social disinhibition, anxiety, and attention problems (Klein-Tasman & Mervis, 2003; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006).

The Child Behavior Checklist (CBCL; Achenbach, 1991; Achenbach & Rescorla, 2000, 2001) is the most commonly used measure for assessing behavioral and emotional problems in children with WS. The 11 published studies of the CBCL include six based on United States (US) samples (Dilts, Morris, & Leonard, 1990; Fidler, Hodapp, & Dykens, 2000; Fu, 2012 – also reported in Fu, Lincoln, Bellugi, & Searcy, 2015; Greer, Brown, Pai, Choudry, & Klein, 1997; Klein-Tasman, Lira, Li-Barber, Gallo, & Brei, 2015; Pagon, Bennett, LaVeck, Stewart, & Johnson, 1987), and one study each from Australia (Porter, Dodd, & Cairns, 2009); Brazil (Teixeira, Monteiro, Velloso, Kim, & Carreiro, 2010), Germany (Sarimski, 1997), Greece (Papaeliou et al., 2012), and Spain (Pérez-García, Granero, Gallastegui, Pérez-Jurado, & Brun-Gasca, 2011). Most of these studies used the CBCL 4–18 (Achenbach, 1991), which included eight empirically-derived problem (“syndrome”) scales based on factor analysis of parental responses to individual items. The more-recent CBCL 6–18 (Achenbach & Rescorla, 2001) and/or CBCL 1½-5 (Achenbach & Rescorla, 2000), which provide both empirically-based scales and DSM-oriented scales, were used by Porter et al. (2009), Teixeira et al. (2010), Papaeliou et al. (2012), and Klein-Tasman et al. (2015).

The factor structure of the CBCL, which has been translated into more than 50 languages, has been shown to be consistent for children in the general population across a wide variety of societies. In particular, the eight-syndrome model of the CBCL derived from the original factor analysis of parental responses to the US version of the CBCL 4–18 fit the data from each of the 42 societies considered by Ivanova et al. (2007) or Rescorla et al. (2012). Furthermore, the items that received high, medium, or low rates of endorsement within a society were consistent across societies. At the same time, Ivanova et al. (2007) and Rescorla et al. (2012) found significant cross-cultural differences in Total Problems raw score, with means ranging from 13.1 (Japan) to 40.8 (Brazil). The US Total Problems raw score (23.74) was very close to the 42-society mean (24.04, SD = 6.74).

These cross-national comparisons were based on children in the general population. Until now no cross-national studies focused on the problem behaviors of children with genetic syndromes have been reported. Such comparisons are important because they allow for consideration of possible cultural influences on either the pattern or intensity of behavioral problems in children from different countries who share the same genetic basis for their developmental disability. The purpose of the present study was to provide a cross-national comparison of children with a specific genetic syndrome, WS, from Spain and the US, two occidental countries on different continents.

Prior Child Behavior Checklist Findings: Individuals with WS

Table 1 lists the 10 studies of individuals with WS whose authors reported descriptive statistics (proportion of participants who scored in the borderline/clinical range and/or mean or median T-score) for at least one of the CBCL scales or higher-order factors. All of these

studies were cross-sectional. Studies are ordered by the approximate mean chronological age (CA) of the sample, which ranged from 5.13 years for Papaeliou et al. (2012) and 6.13 years for Fidler et al. (2000) to 18.16 years for Porter et al. (2009) and 28.23 years for Fu (2012; Fu et al., 2015). Note that although the last three studies in the table included adults in their samples, the oldest age for which the CBCL is normed is 18 years. This table provides a comparison of the proportion of individuals with WS who scored in the CBCL's borderline or clinical range relative to the expected proportion based on the norming sample, for each of the scales or higher-order factors for which the authors provided data. Based on the CBCL 6-18 and CBCL 1½-5 T-score and percentile criteria for the borderline and clinical ranges for the empirical and problem scales, 7% of a sample would be expected to score in these ranges (5% borderline, 2% clinical). The criteria used to classify the proportion of participants who scored in the borderline/clinical range as typical, high, very high, or extremely high relative to the norming sample are listed in the table. Basically, the proportion of participants with elevated T-scores (≥ 65) for a particular scale was considered "typical" if it was less than 15% (approximately twice as high as the proportion for the norming sample), "high" if it was between 15% and 29% (approximately three times as high as the proportion for the norming sample), "very high" if it was between 30% and 49%, and "extremely high" if it was 50% or higher. The T-scores reported by Papaeliou et al. (2012) were based on the Greek norms; the T-scores for the remaining studies were based on the US norms.

Table 1 clearly shows that elevated levels of problem behaviors are more common among individuals with WS than among individuals in the general population. The proportion of individuals with elevated T-scores for Total Problems was 30% or higher in every study except the one with the youngest participants (Papaeliou et al., 2012) – which also was the only one of the non-US studies that used country-specific norms – and the level was higher than expected even in that study. The proportion of participants with elevated T-scores for the Internalizing and Externalizing Problems higher-order factors was higher than expected in nine of the 10 studies.

An examination of the proportion of elevated T-scores for the problem scales, however, indicated that the likelihood of elevations was not uniform across the problem scales. In particular, the proportion of participants with scores in the borderline/clinical range was higher than expected for every study that reported descriptive data for the Attention Problems/Attention Deficit Hyperactivity Problems, Thought Problems, and Social Problems scales. Pagon et al. (1987), whose sample included 10 individuals aged 10 – 20 years, did not report descriptive data for any of the CBCL scales or factors. The authors did list the six items that were endorsed by at least 70% of parents; four of the six were on these three scales. Fidler et al. (2000), who reported descriptive data only for the higher-order factors, indicated the four most frequently endorsed items; two of these were on the Attention Problems scale. In addition, the proportion of participants with scores in the borderline/clinical range on the Anxious/Depressed scale (and/or the Anxiety Problems scale) was higher than expected for all but one study.

In strong contrast, the proportion of participants with elevated scores on Rule-Breaking Behavior/Conduct Problems was in the typical range for all but one study for which

descriptive data for these scales were provided. Even for this study (Teixeira et al., 2010), the proportion was elevated only for the empirically-derived scale (Rule-Breaking Behavior); it was in the typical range for the related DSM-oriented scale (Conduct Problems). It also is important to note that this study was conducted in Brazil, the country for which the highest overall Total Problems raw score was reported for children in the general population (Rescorla et al., 2012). Thus, it is possible that based on the Brazilian norms (which had not been published in 2010) the proportion of children with WS who evidenced problems on the Rule-Breaking scale would not have been considered elevated.

Comparisons between CBCL problem behavior T-scores and other variables were examined in some of the prior studies of individuals with WS, with only a few significant relations identified. Correlations between CBCL T-scores and CA were examined in four studies. Fidler et al. (2000), who studied relatively young children, found significant positive correlations with both Internalizing Problems T-score and Externalizing Problems T-score. Klein-Tasman et al. (2015), who studied children and adolescents, found only one significant correlation – a positive correlation with Anxious/Depressed T-score. Porter et al. (2009), who studied primarily adolescents and adults, also found only one significant correlation – a negative correlation with Externalizing Problems T-score. Fu (2012), who studied primarily adults, did not report any significant correlations with CA.

The possibility of gender differences in T-scores was considered in three studies. Pérez-García et al. (2011) and Klein-Tasman et al. (2015) did not find any significant differences as a function of gender. Porter et al. (2009) reported that females had significantly higher T-scores than males for Total Problems, Externalizing Problems, Affective Problems, Somatic Problems, and Conduct Problems. Note, however, that although the parents of all of the participants in this study completed the CBCL, many of the participants were adults rather than children (mean CA: 18.16 years). It is unknown whether significant differences would have been found if the Adult Behavior Checklist (ABCL; Achenbach & Rescorla, 2003) had been used for these participants.

Possible relations between CBCL T-scores and IQ were considered in three studies (Klein-Tasman et al., 2015; Pérez-García et al., 2011; Porter et al., 2009). No significant correlations with parental ratings were found, although Klein-Tasman et al. did report a significant negative correlation with teachers' ratings on the Thought Problems scale. In the only study that considered the relation between CBCL T-scores and adaptive behavior standard scores (Fu, 2012), no significant correlations were reported.

The Present Study

As indicated in the review of prior studies, the proportion of parents who report significant levels of problem behaviors for their child with WS is considerably greater than expected based on the CBCL norming sample. Similar types of problems have been identified not only across the US samples but also across the samples from other countries, including the sample from the prior study conducted in Spain. In most cases, CBCL T-scores have appeared to be independent of CA or IQ, and only a few gender differences have been found. At the same time, there has not been a direct comparison of the levels or types of problem

behaviors of children with WS from different countries, and with the exception of Papaeliou et al. (2012), non-US samples have been scored on US norms rather than country-specific norms. Furthermore, most of the sample sizes have been small enough that the lack of relations between CBCL T-scores and CA or IQ and the infrequency of gender differences could be due to limited power. In the present study, we provide a direct comparison of parental ratings of same-aged children with WS from Spain and the US on the CBCL 6–18. This cross-national approach allows us to identify similarities and differences in pattern and/or intensity of behavioral or emotional problems in children who have the same genetic syndrome but live in different Western cultures. Both raw-score comparisons (equivalent to using the same norms for both groups) and T-score comparisons based on country-specific norms are reported. The Spanish sample included more participants than any previous CBCL study of children with WS and the US sample was almost three times as large as the largest previous child sample.

Method

Participants

Participants were children with WS from two different countries: Spain and the US. All participants had a genetically-determined classic deletion of the WS region (26 – 28 genes on chromosome 7q11.23).

The Spanish sample included 53 children (22 girls, 31 boys) aged 6.00 – 14.18 years (mean: 9.79 years, median: 9.74; SD: 2.80). All participants were recruited from the WS clinics at the Hospitals del Mar and Vall d'Hebron, Barcelona, and/or referred by the Spanish Williams Syndrome Association. The racial/ethnic composition was White Hispanic for 52 participants (98.1%) and biracial for 1 (1.9%). Mean Full Scale IQ ($n = 36$) on the Spanish versions of the Wechsler Intelligence Scale for Children-Revised (WISC-R; Wechsler, 2001) was 54.64 (SD: 12.62, range: 40 – 96). Mean Verbal IQ was 65.28 (SD: 14.97, range: 44 – 116) and mean Performance IQ was 52.69 (SD: 8.55, range: 40 – 74).

The American sample included 145 children (75 girls, 70 boys) aged 6.01 – 14.18 years (mean: 9.69 years, median: 9.25, SD: 2.57). The parents of some participants completed the CBCL 6–18 as part of a longitudinal study. For these participants, data from the most recent CBCL 6–18 administration at which the child was < 14.2 years old were used. The participants' racial/ethnic background was: 112 White non-Hispanic (77.2%), 8 White Hispanic (5.5%), 5 Asian non-Hispanic (3.4%), 4 African American non-Hispanic (2.8%), 12 biracial or tri-racial non-Hispanic (8.3%), and 4 biracial or tri-racial Hispanic (2.8%). Mean Differential Ability Scales-II (Elliott, 2007) General Conceptual Ability standard score (GCA; similar to IQ) was 65.20 (SD: 13.26, range: 33 – 96). Mean standard scores for the three core clusters were 74.46 (SD: 17.00, range: 30 – 106) for Verbal, 78.59 (SD: 14.06, range: 43 – 113) for Nonverbal Reasoning, and 55.15 (SD: 13.77, range: 32 – 85) for Spatial.

The Spanish and American groups were well matched for CA (Mann Whitney U test, $Z = 0.06$, $p = .95$). As intellectual abilities were measured by different assessments for the two groups, a direct comparison of IQ scores was not appropriate. The descriptive statistics for

intellectual abilities for each group were consistent with those typically reported for children with WS assessed by that test.

Measure

The Child Behavior Checklist for Ages 6–18 (CBCL; Achenbach & Rescorla, 2001) provides a parent-report measure of children's behavioral and emotional problems for the past six months. Parents are asked to rate 113 items on a 3-point Likert scale (0 = not true, 1 = somewhat or sometimes true, and 2 = very true or often true). The CBCL includes eight empirically based scales and six scales based on the Diagnostic and Statistical Manual of Mental Disorders IV (DSM-IV). For each scale raw scores are computed and then converted to T scores taking into account gender and age (divided in 2 groups: 6 – 11 years, 12 – 18 years). T scores of 64 or lower are considered to be in the normal range, T scores of 65 – 69 are in the borderline range, and T scores of 70 or higher are in the clinical range. Additionally, the empirically-based scales are grouped into three higher-order factor scales: Internalizing Problems (Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints), Externalizing Problems (Rule-breaking Behavior, Aggressive Behavior) and Total Problems (all items).

T scores for the American sample were based on the norms in Achenbach and Rescorla (2001). As reported by Achenbach and Rescorla, test-retest correlations over a 1-week interval ranged from .82 – .92 for the empirically based scales, from .91 – .94 for the higher-order factors (Internalizing, Externalizing, Total Problems) and from .80 – .93 for the DSM-oriented scales. Cronbach's *alpha* ranged from .78 – .94 for the empirically-based scales, from .90 – .97 for the higher-order factors, and from .72 – .91 for the DSM-oriented scales. Mean Total Problems raw score was 23.74, which is very close to the mean of 24.04 (SD: 6.74) reported by Rescorla et al. (2012) for 42 societies.

T scores for the Spanish sample were based on the provisional normative data available for Spain (Unitat d'Epidemiologia i de Diagnòstic en Psicopatologia del Desenvolupament, 2013). The provisional norming sample included 1430 children aged 6 – 17 years (715 girls, 715 boys). Separate norms were generated for two age groups: 6 – 11 years and 12 – 17 years, with different norms for girls and boys in each age group. Bernedo, Fuentes, and Fernández (2008) reported that Cronbach's *alpha* values ranged from .88 – .93 for the higher-order factors, based on a sample of 68 children. Lacalle, Ezpeleta, and Doménech (2012) reported Cronbach's *alpha* values for the DSM-oriented scores, based on a sample of 420 clinically-referred children. Values were in the moderate to good range (.70 – .82) for all of the scales except Anxiety (.59). The mean Total Problems raw score in the sample on which the provisional norms were based (Bernedo et al., 2008) was 29.7, which is 0.84 SD above the mean for the 42 societies reported in Rescorla et al. (2012). As the T-score range for the problem scales is 50 – 100 for the US norms, we set the lowest T-score for the problem scales for the Spanish sample at 50 and the highest T-score at 100 so that the range would be the same for the two countries.

Procedure

Parents completed the CBCL following the standardized procedures, as part of a larger assessment.

Data Analysis

The data for several variables were not normally distributed. In addition, the American sample was almost three times as large as the Spanish sample. For these reasons, nonparametric statistics were used for all analyses. Mann-Whitney U tests were used for between-country comparisons of CBCL raw scores and T scores, and Chi square tests were used for between-country comparisons of proportions. The correlation coefficient r was used to determine effect sizes for the Mann-Whitney U and Chi-square tests, with .1 indicating a small effect, .3 indicating a medium effect, and .5 indicating a large effect. Spearman rank-order correlations were computed to examine relations between CBCL T scores and CA or IQ, separately for each nationality group. To adjust for multiple comparisons, α was set at .01.

Results

Preliminary analyses indicated no significant gender differences on any of the CBCL scales or higher-order factors for children from either Spain or the US. Accordingly, gender was not included as a factor in any of the analyses reported below.

Raw Score and T-Score Comparisons

Descriptive statistics for raw scores and T-scores for the CBCL empirically-based scales and higher-order factors are presented in Table 2. To determine if there were significant differences in the distributions of scores as a function of nationality, Mann Whitney U tests were conducted. As indicated in the table, there were significant differences in raw scores between the Spanish and American groups on four of the eight empirically-based scales: Anxious/Depressed ($Z = 2.77, p = .006, r = .20$), Withdrawn/Depressed ($Z = 3.22, p = .001, r = .23$), Somatic Complaints ($Z = 4.77, p < .001, r = .34$), and Social Problems ($Z = 3.28, p = .001, r = .23$). There also were significant differences in raw scores for all three higher-order factors (Internalizing Problems: $Z = 4.53, p < .001, r = .32$; Externalizing Problems: $Z = 2.67, p = .007, r = .19$; Total Problems: $Z = 3.51, p < .001, r = .25$). In all cases, the distributions were higher (indicating more difficulty) for the Spanish sample than for the American sample. In contrast, a very different pattern emerged when between-nationality comparisons were based on country-specific norms. In particular, the T-score distributions did not differ significantly on any of the higher-order factors and differed significantly on only one empirically-based scale – Social Problems ($Z = 2.60, p = .009, r = .18$).

Descriptive statistics for the DSM-oriented scales are provided in Table 3. Mann-Whitney U tests indicated a significantly higher distribution of raw scores for the Spanish group than for the American group for Affective Problems ($Z = 4.55, p < .001, r = .32$), Anxiety Problems ($Z = 3.35, p = .001, r = .24$), and Somatic Problems ($Z = 2.82, p = .005, r = .16$). In contrast, T-score comparisons using country-specific norms indicated only one significant difference; the distribution for the American group was significantly higher than for the Spanish group

for Conduct Problems ($Z = -2.86$, $p = .004$, $r = .20$). Note, however, that the means and medians for both groups were well within the normal range on this scale.

In order to further investigate the significant between-group difference in T-scores on the Social Problems scale, an item analysis was conducted. The items included on this scale, along with the distribution of raw scores for each item as a function of country, are listed in Table 4. Chi-square tests indicated that three items were endorsed (score of 1 or 2) by a significantly higher percentage of parents of the Spanish group than the American group: Clings to adults or too dependent ($\chi^2 = 31.26$, $p < .001$, $r = .40$), Gets teased a lot ($\chi^2 = 21.85$, $p < .001$, $r = .33$), and Poorly coordinated or clumsy ($\chi^2 = 7.89$, $p = .005$, $r = .20$). One item was endorsed by a significantly higher percentage of parents of the American group than the Spanish group: Speech problem ($\chi^2 = 8.70$, $p = .003$, $r = .21$).

Categorical Comparisons: Scales and Higher-Order Factors

The percentage of children in each group who scored in the normal, borderline, or clinical range on each of the CBCL scales and higher-order factors is indicated in Table 5. For this table the same criteria (listed in Table 1) for determining the categorical assignment of a particular T score were used for both the scales and the higher-order factors. As shown in the table, the most common problem area was attention, with more than half of the sample from each country scoring in the borderline or clinical range on the empirically-based Attention Problems scale and the DSM-oriented ADH Problems scale. Chi square tests were used to determine if the proportion of children who scored in the normal vs. the borderline or clinical range on each scale or factor differed as a function of nationality. The only significant difference was for the Social Problems scale ($\chi^2 = 6.95$, $p = .008$, $r = .19$), for which a significantly higher proportion of the Spanish sample than the American sample scored in the borderline or clinical range.

Relations between CBCL T-scores and CA or IQ

To determine if children's CBCL T-scores on the empirically-based scales, DSM-oriented scales, or higher-order factors were related to their CA, Spearman correlations were conducted, separately for each nationality. For the Spanish sample, the only significant correlation was with Internalizing Problems T ($r_s = .36$, $p = .009$), indicating that internalizing problems increased as CA increased. For the American sample, none of the correlations was significant.

Another series of Spearman correlations was conducted, separately for each country, to determine possible relations between children's CBCL T-scores and IQ. For the Spanish sample, none of the correlations was significant. For the American sample, the correlations with Thought Problems T-score ($r_s = -.27$, $p = .001$), Attention Problems T-score ($r_s = -.25$, $p = .003$), Anxiety Problems T-score ($r_s = -.28$, $p = .001$), and ADH Problems T-score ($r_s = -.22$, $p = .009$) were significant. In each of these cases, higher T-scores were associated with lower IQ. The effect size for each of these correlations is small.

Discussion

The findings from this study revealed both differences and similarities in parental reports of problem behaviors in children with WS from Spain and from the US. At the absolute level (as measured by raw scores), the score distribution for the Spanish sample was significantly higher (indicating significantly greater problem behavior) than for the American sample for all of the higher-order factors, four of the eight empirically-derived scales, and three of the six DSM-oriented scales. Thus, parents of Spanish children with WS reported considerably higher levels of behavioral and emotional problems for their children than did parents of American children with WS. However, at the relative level (as measured by T-scores derived from country-specific norms), almost all of these differences disappeared. In the remainder of the Discussion, we first consider this pattern of findings in relation to prior cross-national research on other aspects of the WS phenotype. Next, we compare the present findings to prior CBCL findings from studies of children with WS. We also compare the present categorical classifications based on T-scores on the CBCL DSM-oriented scales to diagnostic findings from prior studies that used DSM-based clinical interviews with parents of children with WS. We then consider possible reasons for the significant T-score and categorical classification differences between nationalities for the Social Problems scale. Finally, we consider limitations of this study and directions for future research.

Comparisons with Prior Cross-National Studies of Children with WS Addressing Other Aspects of Development

In the present study, we found significant cross-national differences in parental reports of absolute levels of behavioral problems for their child with WS for half of the CBCL scales, with children from Spain scoring significantly higher than children from the US. However, once the behavior problems of the children with WS were compared to those of children in the general population from their own country, almost all of the cross-national differences disappeared; the only meaningful significant cross-national difference that remained was for the Social Problems scale. This pattern of significant cross-cultural differences between groups of children with WS from different countries in absolute levels of specific behaviors accompanied by cross-cultural similarities between WS groups in relative levels of the same behaviors in comparison to their compatriots in the general population is consistent with that reported in two prior cross-national comparisons of children with WS focused on other components of social behavior. In the first study, based on parental ratings from the Salk Institute Sociability Questionnaire (Jones, Bellugi, Lai, Chiles, Reilly, & Lincoln, 2000), Zitzer-Comfort, Doyle, Masataka, Korenberg, and Bellugi (2007) found that children with WS in the US were significantly more likely to approach strangers than were children with WS in Japan. However, within each nationality, the children with WS scored significantly higher than the TD children. In the second study, Reilly, Bernicot, Vicari, Lacroix, and Bellugi (2003) reported that the narratives of Italian children with WS included a higher proportion of propositions containing socially expressive language than did those of American children with WS, whose narratives included a higher proportion than did those of French children with WS. At the same time, within each nationality the WS group produced a considerably higher proportion of propositions containing socially expressive language than did the matched TD group. Taken together, these findings suggest that there are cross-

national differences in absolute levels of particular types of behavior associated with the WS phenotype (e.g., inappropriate social approach) that are likely driven by cultural differences. At the same time, when the comparison is to children in the general population from the same culture, clear cross-national similarities emerge for children with WS, suggesting an important role for genetics in the behavioral phenotype of WS, transcending cultural differences.

Relations to Prior CBCL Findings for Children with WS

In prior studies of the performance of children with WS on the CBCL, the scales on which difficulties were most likely to be identified were Attention Problems/ADH Problems and Social Problems; for all of the studies for which the mean age of participants was < 16 years, the proportion of children with clear difficulty was either very high or extremely high on all three scales (Table 1). Problems also were very likely to be noted on the Thought Problems scale and the Anxious-Depressed/Anxiety Problems scales. The scales for which children were least likely to evidence significant difficulties were Rule-Breaking Behavior/Conduct Problems. The findings from the present study were very similar to this pattern. For the Spanish sample, the proportion of participants identified as having difficulties was extremely high for the Attention scales, very high for Social Problems, Thought Problems, and Anxiety Problems, and typical for the Conduct scales. For the US sample, the pattern was almost identical: extremely high for the Attention scales, very high for Thought Problems and Anxiety Problems, at the top of the high range for Social Problems, and typical for the Conduct scales.

The possibility of significant relations between CBCL T-scores and gender, CA, or IQ also was addressed. In the present study, no significant gender differences in CBCL T-scores were identified for either the Spanish group or the US group. This finding is consistent with those from the two previous examinations of CBCL T-scores for samples of individuals with WS for which all or most of participants were 18 years or younger that considered the possibility of gender differences (Klein-Tasman et al., 2015; Pérez-García et al., 2011). Across the two groups included in the present study, only one significant relation with CA was found: For the Spanish sample, Internalizing Problems T-score was significantly correlated with CA. This finding also was reported in one of the previous studies (Fidler et al., 2000). Overall, the dearth of significant relations with CA in the present study was consistent with the rarity of significant relations involving CA in previous studies. The failure to find additional significant relations with CA in the present study is noteworthy given the large number of children included in the US sample ($n = 145$).

Previous studies have not reported any significant correlations between CBCL T-scores based on parent ratings and IQ, although a significant negative correlation between CBCL Thought Problems T-score based on teacher ratings and IQ was found (Klein-Tasman et al., 2015). In the present study, no significant relations between CBCL T-scores and IQ were found for the Spanish sample. However, the previous finding of a significant negative relation between CBCL Thought Problems T-score and IQ was replicated for the US sample. In addition, significant negative correlations between CBCL T-score and IQ were found for the US sample for the Anxiety Problems, ADH Problems, and Attention Problems scales.

All of these correlations represented small effect sizes. It is possible that the failure to find significant correlations with IQ in previous studies and in the Spanish sample in the present study is due in part to a lack of power; the US sample in the present study is almost 3 times as large as any of the other samples.

Relations between CBCL DSM-oriented Scale Categorical Classifications and DSM-IV Diagnoses

In the present study, the DSM-oriented scale for which the most children were categorized in the borderline or clinical range was ADH Problems; for both nationalities, fewer than half of the children scored in the normal range. The scale for which the next largest proportion of children with WS was classified in the borderline or clinical range was Anxiety Problems. In contrast, only a small proportion of the children in each group was classified in the borderline or clinical range for Oppositional Defiant Problems or for Conduct Problems. This pattern is consistent with what has been reported in studies of children with WS that have used clinical interviews such as the Anxiety Disorders Interview Schedule-Parent (ADIS-P; Silverman & Albano, 1996) to identify possible DSM-IV diagnoses. In the largest of these studies, Leyfer et al. (2006) used the ADIS-P to identify DSM-IV diagnoses for 119 children with WS aged 4 – 16 years. Prevalence rates were 64% for ADHD and 54% for specific phobia (by far the most common anxiety disorder diagnosis for children with WS) but only 3% for Oppositional Defiant Disorder and 0% for Conduct Disorder. Kennedy, Kaye, and Sadler (2006) used the ADIS-P to determine DSM-IV diagnoses for a somewhat older sample of 21 individuals with WS (mean CA = 16 years). The findings were similar: 48% had at least one anxiety diagnosis and 43% had a diagnosis of ADHD but only 5% had a diagnosis of Oppositional Defiant Disorder and 0% had a diagnosis of Conduct Disorder.

Possible Bases for Significant Cross-cultural Differences in CBCL Social Problems T-score Distribution

Even after using nationality-specific norms, the mean CBCL Social Problems T-score was significantly higher for the Spanish WS group than for the American WS group and a significantly higher proportion of the Spanish group than the American group scored in the borderline or clinical range on this scale. Relative to the parents of the American group, the parents of the Spanish group were significantly more likely to endorse items related to being teased, clinging to adults or being too dependent on them, and being poorly coordinated or clumsy. The parents of the American group endorsed the speech problem item significantly more often than did the parents of the Spanish group.

These Social Problems differences are likely due to an interaction between cultural differences between Spain and the US and genetically-based characteristics of individuals with WS. Whereas Anglo-American culture typically is characterized as individualistic, Spanish culture is characterized as collectivistic. Thus, relative to Anglo-American culture, Spanish culture puts less emphasis on independence and individual goals and more on interdependence and shaping one's behavior to conform to in-group norms to promote relationships that are smooth and harmonious (Benet-Martínez & John, 2000; Triandis, 2001). In Spain, as in other collectivist cultures, adolescents are considerably more likely to spend free time with their peers than are adolescents in the US (Currie et al., 2008). Children

with WS, although highly motivated to interact with other people, have social skills deficits, including excessive approach toward others and difficulties with social adjustment or understanding (e.g., Davies, Udwin, & Howlin, 1998). These behaviors lead to social vulnerability (Jawaid et al., 2012), including victimization by teasing (Fisher, Moskowitz, & Hodapp, 2013), problems that are most likely to occur in unstructured settings with peers. For cultural reasons, Spanish individuals with WS typically spend considerably more time than American individuals with WS in these types of settings, leading to their parents' heightened awareness of the difficulties highlighted by these situations, including being teased and being too dependent on other people.

Individuals with WS have difficulty with balance and coordination (Pober, 2010) and their speech and language skills are delayed relative to their CA (Garayzabal & Cuetos, 2010; Mervis & Velleman, 2011). However, there is no evidence of cross-national differences either in motor coordination or in speech/language abilities for individuals with WS. It is possible that the significant differences in parental ratings on items related to these characteristics reflect differences in the value that parents in different cultures place on motor abilities relative to speech abilities. Parental expectations are influenced by cultural models (Suizzo, 2007) and the meaning of disability is culturally relative (Neely-Barnes & Marcenko, 2004), so differences in the relative weight parents give to their children's motor and speech abilities could occur. We are not aware of any research that addresses this possibility, however.

Limitations and Future Directions

The data reported in this study are parental ratings. No ratings were collected from other individuals who spent time with the participants with WS (e.g., teachers). In the only study of children with WS that compared parental and teacher ratings (Klein-Tasman et al., 2015), both similarities and differences were found. In particular, both parents and teachers commonly gave the children high ratings on attention problems, anxiety problems, social problems, and thought problems. However, parents rated children higher (worse) than did teachers on attention problems and affective problems whereas teachers rated children higher than did parents on oppositionality. To further understand cross-cultural similarities and differences in the behavior of children with WS across multiple settings, comparisons of parental and teacher ratings would be valuable. Comparisons of maternal and paternal ratings also would be useful.

The present study did not include either direct observation of the participants or elicitation of descriptions from the parents of how their child would behave in a particular situation. Without this information, the possibility remains that the significantly higher raw scores found for the Spanish group relative to the American group on half of the CBCL scales reflect at least in part differences in parental interpretation of a particular behavior as problematic (perhaps due to cultural beliefs) rather than differences in actual behavior. We mentioned this possibility with regard to significant differences between the two groups of parents in the proportion endorsing motor coordination problems or speech problems, given that there are no known differences in these abilities for children with WS as a function of nationality. This possibility also was raised previously by Zitzer-Comfort et al. (2007), who

reported that when parents provided written descriptions of their children's social approach behavior, the descriptions for Japanese and American children appeared to be more similar than would have been expected given the significant differences in parental ratings of the children's social approach behavior. Future research in which parental descriptions of behavior in specific situations are elicited and then scored by raters blind to the children's nationality would help to address this possibility.

Finally, the present study included only one parental questionnaire (CBCL 6-18) and only two nationalities (Spain and the US), and all of the children had the same genetic syndrome. A more complete understanding of parental perceptions of similarities and differences in the behavioral phenotype of children with WS as a function of culture will require comparisons based on additional questionnaires measuring other types of behavior across multiple cultures for not only school-aged children but also younger children. These types of studies are considerably easier to conduct with the ASEBA measures (www.aseba.org), including the CBCL 6-18, for which culture-specific norms or culture-group specific norms are available. Development of these types of norms for other commonly used questionnaire assessments would greatly facilitate cross-cultural research both on TD children and on children with developmental disabilities. And consideration of children with other types of genetic syndromes would allow for determination of similarities and differences across not only nationalities but also genetic disorders.

In conclusion, the present study contributes to the understanding of patterns of similarities and differences in the behavior of children with WS from different countries. At the absolute level, we found that the Spanish group of children with WS was rated as evidencing significantly greater problem behavior than the American group of children with WS both overall and on half of the empirically-based scales and DSM-based CBCL scales. However, almost all of these cross-national differences disappeared once the children with WS were compared to country-specific norms, indicating that children with WS from Spain and the US showed similar patterns of problem behaviors relative to TD children of the same nationality. These findings, which are consistent with those of prior cross-national comparisons of children with WS addressing other aspects of the WS phenotype, provide further evidence that although there are absolute differences in the performance of children with WS as a function of cultural conventions (which often vary as a function of nationality), comparisons of children with WS relative to compatriot TD children identify patterns that are largely consistent across nationalities, reflecting the genetic component of WS.

Acknowledgments

Débora Pérez-García was supported by a predoctoral fellowship from Instituto de Salud Carlos III (FI11/00656). Data collection for the Spanish sample was supported by grants from the Spanish Ministries of Science, Economy & Competitiveness (SAF04/6382 & PI13/02481) and the European Community (EC-FP6-37627) to Luis A. Pérez-Jurado. Data collection for the US sample was supported by grants from the National Institute of Child Health and Human Development (R37 HD29957) and the National Institute of Neurological Disorders and Stroke (R01 NS35102) to Carolyn Mervis. Data analysis and manuscript preparation were supported by NICHD grant R37 HD29957 and grant WSA 0104 from the Williams Syndrome Association to Carolyn Mervis. We would like to thank the children and parents who participated in this research. We also thank the members of the Neurodevelopmental Sciences Laboratory at the University of Louisville who collected or entered the US data for this project.

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Literature Summary: Proportion of Children with WS who have Borderline/Clinical Levels of CBCL Problem Behaviors, Relative to Proportion of Norming Sample

Table 1

Study First Author (N, CA Range)	Country/ Norm Reference Group	Scale Type	Total Problems	Internalizing Problems	Externalizing Problems	Anxious/ Depressed (Anxiety Problems)	Withdrawn/ Depressed (Affective Problems)	Somatic Complaints (Somatic Problems)	Social Problem s	Thought Problems	Attention Problems (Att. Deficit Hyperactivity Problems)	Rule- Breaking Behavior (Conduct Problems)	A aggressive Behavior (Oppositional Defiant Problems)
Papaeliou (20, 3–6 yrs)	Greece/ Greece	Emp	high	typical	high		high	high			very high		high
Fidler (20, 3–10 yrs)	USA/ USA	Emp	ext. high	high	very high								
Dilts (48, 4–16 yrs)	USA/ USA	Emp									ext. high		
Teixeira (10, 5–16 yrs)	Brazil/ USA	Emp	very high	very high	very high	very high	very high	high	very high	very high	very high	high	typical
		DSM				very high	very high	high			very high	typical	typical
Sarimski (14, 4–18 yrs)	Germany/ USA	Emp	ext. high										
Greer (15, 4–18 yrs)	USA/ USA	Emp	very high	very high	very high	typical		typical	ext. high	ext. high	ext. high	typical	
Klein-Tasman (52, 6-17 yrs)	USA/ USA	Emp	very high	high	high	high	typical	high	very high	very high	ext. high	typical	high
		DSM				very high	high	high			very high	typical	high
Pérez-García (25, 5–27 yrs)	Spain/ USA	Emp	ext. high	ext. high	high	ext. high	high	very high	very high	high	ext. high	typical	very high
Porter (31, 6–48 yrs)	Australia/ USA	Emp	very high	high	typical								
		DSM				very high	typical	high			high	typical	typical
Fu (91, 12–53 yrs)	USA/ USA	Emp	very high	very high	very high	high							

Abbreviations: Att. = Attention, Emp = empirically-determined scales, DSM = DSM-oriented scales, ext. = extremely, yrs = years

Note: T ranges: Typical = 64, Borderline = 65 – 69, Clinical = 70. Scales listed in parentheses are DSM-oriented. Based on these criteria, 5% of the norming sample scored in the Borderline range and 2% scored in the clinical range on each of the problem scales. Criteria: “Typical:” < 15% scored in the Borderline or Clinical range. If the % who scored in the Borderline or Clinical range was not provided, the following criteria were used: (mean T-score + 1 SD) was in the normal range, or boxplot suggested that < 15% scored in the Borderline or Clinical range. “High:” 15% but < 30% scored in the Borderline or Clinical range, or (mean T-score + 1 SD) was in the borderline range, or boxplot suggested that 15% but < 30% scored in the Borderline or Clinical range. “Very high:” 30% but < 50% scored in the Borderline or Clinical range, or (mean T-score + 1 SD) was in the clinical range, or boxplot suggested that 30% but less than 50% scored in the Borderline or Clinical range. “Extremely high:” 50% scored in the Borderline or Clinical range, or mean or median T-score was in the Clinical range.

Table 2
Descriptive Statistics for Raw and T Scores on CBCL Empirically Based Scales and Higher-Order Factors as a Function of Country

	Raw scores						T scores							
	Spain (N = 53)			USA (N = 145)			p	Spain (N = 53)			USA (N = 145)			p
	Mean (SD)	Median	Range	Mean (SD)	Median	Range		Mean (SD)	Median	Range	Mean (SD)	Median	Range	
Anxious/ Depressed	5.96 (4.02)	5.00	0 – 17	4.10 (2.99)	4.00	0 – 13	.006*	57.79 (9.75)	52.00	50 – 80	56.46 (6.36)	54.00	50 – 74	.652
Withdrawn/ Depressed	2.57 (2.15)	2.00	0 – 7	1.50 (1.66)	1.00	0 – 8	.001*	56.94 (8.22)	53.00	50 – 81	54.58 (5.25)	54.00	50 – 70	.273
Somatic Complaints	4.02 (2.98)	4.00	0 – 14	1.94 (2.03)	1.00	0 – 8	.000*	59.43 (10.74)	58.00	50 – 100	56.25 (6.33)	53.00	50 – 74	.211
Social Problems	6.87 (3.26)	7.00	2 – 16	5.11 (3.00)	5.00	0 – 16	.001*	66.49 (12.80)	64.00	50 – 100	60.64 (7.05)	60.00	50 – 86	.009*
Thought Problems	5.21 (3.10)	4.00	1 – 13	5.07 (3.16)	4.00	0 – 16	.792	64.11 (14.55)	60.00	50 – 100	63.60 (7.77)	64.00	50 – 82	.148
Attention Problems	10.91 (3.13)	12.00	3 – 16	10.17 (3.29)	11.00	2 – 17	.115	68.15 (8.84)	69.00	50 – 87	67.63 (7.78)	67.00	51 – 90	.280
Rule-breaking Behavior	3.40 (2.16)	3.00	0 – 10	2.66 (2.00)	2.00	0 – 12	.013	56.19 (7.63)	53.00	50 – 77	56.28 (5.70)	55.00	50 – 72	.136
Aggressive Behavior	9.42 (5.97)	9.00	0 – 29	7.21 (5.32)	6.00	0 – 23	.014	58.58 (9.90)	57.00	50 – 94	57.70 (7.37)	56.00	50 – 81	.768
Internalizing Problems	12.55 (7.14)	11.00	0 – 32	7.54 (5.07)	7.00	0 – 26	.000*	56.87 (10.59)	55.00	38 – 90	54.06 (9.09)	54.00	33 – 72	.267
Externalizing Problems	12.81 (7.46)	12.00	2 – 36	9.86 (7.10)	8.00	0 – 35	.007*	54.79 (9.92)	54.00	41 – 90	55.50 (9.50)	56.00	33 – 76	.232
Total Problems	55.04 (21.39)	54.00	17 – 105	42.63 (18.81)	42.00	4 – 92	.000*	60.26 (9.85)	58.00	43 – 87	60.21 (7.95)	61.00	36 – 75	.679

Note:

* p .010

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Table 3
Descriptive Statistics for Raw and T Scores for DSM-Oriented Scales as a Function of Country

Item	Raw scores						T scores					
	Spain (N = 53)			USA (N = 145)			Spain (N = 53)			USA (N = 145)		
	Mean (SD)	Median	Range	Mean (SD)	Median	Range	Mean (SD)	Median	Range	Mean (SD)	Median	Range
Affective Problems	5.45 (3.31)	5.00	0 – 12	3.12 (2.55)	3.00	0 – 13	61.83 (11.23)	58.00	50 – 87	58.78 (7.04)	59.00	50 – 77
Anxiety Problems	4.87 (2.59)	4.00	0 – 11	3.43 (2.40)	3.00	0 – 10	61.74 (10.73)	58.00	50 – 91	60.32 (9.54)	60.00	50 – 77
Somatic Problems	1.87 (1.98)	1.00	0 – 8	1.08 (1.51)	0.00	0 – 5	55.38 (8.34)	50.00	50 – 84	55.09 (6.57)	50.00	50 – 70
ADH Problems	8.26 (2.51)	9.00	2 – 13	7.89 (2.64)	8.00	1 – 14	66.28 (9.22)	66.00	50 – 88	64.62 (6.60)	66.00	50 – 80
Opp. Def. Problems	3.79 (2.54)	2.00	0 – 12	3.10 (2.34)	3.00	0 – 10	56.60 (8.01)	55.00	50 – 82	56.82 (7.09)	55.00	50 – 80
Conduct Problems	2.94 (2.75)	2.00	0 – 12	2.72 (3.20)	2.00	0 – 18	54.45 (7.37)	50.00	50 – 82	56.52 (7.09)	54.00	50 – 79

Note: Opp. Def. = Oppositional Defiant

*
p .010

Table 4

Distribution of Raw Scores (Number and Percentage of Participants) on Items Included on the Social Problems Scale as a Function of Country

Item	Spain				USA				P Not True vs. Somewhat or Very True
	0 Not True	1 Somewhat or Sometimes True	2 Very True or Often True	0 Not True	1 Somewhat or Sometimes True	2 Very True or Often True	0 Not True	1 Somewhat or Sometimes True	
Clings to adults or too dependent	7 (13%)	22 (41.5%)	24 (45.3%)	84 (57.9%)	44 (30.3%)	17 (11.7%)			.000 *
Complains of loneliness	31 (58.5%)	20 (37.7%)	2 (3.8%)	109 (75.2%)	33 (22.8%)	3 (2.1%)			.022
Doesn't get along with other kids	38 (71.7%)	13 (24.5%)	2 (3.8%)	98 (67.6%)	45 (31.0%)	2 (1.4%)			.581
Easily jealous	34 (64.2%)	14 (26.4%)	5 (9.4%)	100 (69.0%)	40 (27.6%)	5 (3.4%)			.521
Feels others are out to get him/her	43 (81.1%)	9 (17.0%)	1 (1.9%)	134 (92.4%)	11 (7.6%)	0 (0%)			.022
Gets hurt a lot, accident prone	43 (81.1%)	6 (11.3%)	4 (7.5%)	107 (73.8%)	34 (23.4%)	4 (2.8%)			.286
Gets teased a lot	17 (32.1%)	33 (62.3%)	3 (5.7%)	100 (69.0%)	40 (27.6%)	5 (3.4%)			.000 *
Not liked by other kids	33 (62.3%)	17 (32.1%)	3 (5.7%)	102 (70.3%)	37 (25.5%)	6 (4.1%)			.280
Poorly coordinated or clumsy	6 (11.3%)	24 (45.3%)	23 (43.4%)	45 (31.0%)	69 (47.6%)	31 (21.4%)			.005 *
Prefers being with younger kids	16 (30.2%)	28 (52.8%)	9 (17.0%)	67 (46.2%)	56 (38.6%)	22 (15.2%)			.043
Speech problem	31 (58.5%)	12 (22.6%)	10 (18.9%)	51 (35.2%)	42 (29.0%)	52 (35.9%)			.003 *

* p .010

Table 5

Percentage of Participants in the Normal, Borderline, and Clinical Ranges on the CBCL Empirically-Based and DSM-Oriented Scales and the Higher Order Factors as a Function of Country

	Spain			USA			<i>p</i> for Spain – US comparison: % Normal vs. % Borderline or Clinical
	Normal	Borderline	Clinical	Normal	Borderline	Clinical	
Empirically-based scales							
Anxious/Depressed	73.6	7.5	18.9	84.8	11.7	3.4	.069
Withdrawn/Depressed	84.9	5.7	9.4	92.4	6.2	1.4	.112
Somatic complaints	71.7	11.3	17.0	85.5	9.7	4.8	.026
Social problems	50.9	7.5	41.5	71.0	18.6	10.3	.008*
Thought problems	66.0	9.4	24.5	52.4	18.6	29.0	.087
Attention problems	30.2	20.8	49.1	34.5	28.3	37.2	.570
Rule-breaking behavior	86.8	3.8	9.4	90.3	4.1	5.5	.472
Aggressive behavior	83.0	3.8	13.2	80.0	13.1	6.9	.633
DSM-oriented scales							
Affective problems	66.0	9.4	24.5	73.8	17.2	9.0	.283
Anxiety problems	62.3	15.1	22.6	62.1	18.6	19.3	.980
Somatic problems	86.8	5.7	7.5	88.3	6.9	4.8	.777
ADH problems	41.5	22.6	35.8	48.3	29.0	22.8	.398
Oppositional defiant problems	86.8	5.7	7.5	85.5	7.6	6.9	.820
Conduct problems	92.5	1.9	5.7	86.2	8.3	5.5	.233
Higher-level factors							
Internalizing	81.1	1.9	17.0	86.9	12.4	0.7	.222
Externalizing	88.7	5.7	5.7	81.4	10.3	8.3	.310
Total	67.9	15.1	17.0	66.2	17.2	16.6	.820

* *p* .010