

Increased Cross-Gender Identification Independent of Gender Role Behavior in Girls with
Congenital Adrenal Hyperplasia: Results from a Standardized Assessment of 4- to 11-Year-
Old Children

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ABSTRACT

While reports showing a link between prenatal androgen exposure and human gender role behavior are consistent and the effects are robust, associations to gender identity or cross-gender identification are less clear. The aim of the current study was to investigate potential cross-gender identification in girls exposed prenatally to high concentrations of androgens due to classical congenital adrenal hyperplasia (CAH). Assessment included two standardized measures and a short parent interview assessing frequency of behavioral features of cross-gender identification as conceptualized in Part A of the diagnostic criteria for gender identity disorder (GID) in the DSM-IV-TR. Next, because existing measures may have conflated gender role behavior with gender identity and because the distinction is potentially informative, we factor analyzed items from the measures which included both gender identity and gender role items to establish the independence of the two constructs. Participants were 43 girls and 38 boys with CAH and 41 unaffected female and 31 unaffected male relatives, aged 4- to 11-years. Girls with CAH had more cross-gender responses than female controls on all three measures of cross-gender identification as well as on a composite measure of gender identity independent of gender role behavior. Furthermore, parent report indicated that 5/39 (12.8%) of the girls with CAH exhibited cross-gender behavior in all five behavioral domains which comprise the cross-gender identification component of GID compared to 0/105 (0.0%) of the children in the other three groups combined. These data suggest that girls exposed to high concentrations of androgens prenatally are more likely to show cross-gender identification than girls without CAH or boys with and without CAH. Our findings suggest that prenatal androgen exposure could play a role in gender identity development in healthy children, and may be relevant to gender assignment in cases of prenatal hormone disruption, including, in particular, cases of severely virilized 46,XX CAH.

KEY WORDS: Androgens; congenital adrenal hyperplasia (CAH); gender dysphoria; gender identity

1 INTRODUCTION

2 Exposure to androgens during critical periods of fetal development is linked to
3 masculinization and defeminization of human behavior (Cohen-Bendahan, van de Beek, &
4 Berenbaum, 2005; Hines, 2011). A primary source of information on the effects of androgens
5 on human development has been studies of girls and women who have classical congenital
6 adrenal hyperplasia (CAH), which exposes them to abnormally high concentrations of
7 androgens, including testosterone, beginning *in utero* (Merke & Bornstein, 2005). Girls with
8 CAH are typically born with some degree of physical virilization (fused labia, enlarged
9 clitoris) caused by their prenatal androgen exposure and may later also show masculinized
10 gender role behavior. *Gender role behavior* refers to behaviors which, on average, are more
11 typical of one gender or the other and includes preferences for specific toys, playmates, and
12 play styles. As a group, girls with CAH are more likely than other girls to prefer boys' toys,
13 boys as playmates, and masculine play styles. This alteration in childhood play behavior is
14 well-established, with over 10 different studies reporting that girls with CAH show more
15 masculine play behavior compared to unaffected female relatives or matched controls (e.g.,
16 Berenbaum & Hines, 1992; Dittmann et al., 1990; Ehrhardt & Baker, 1974; Ehrhardt,
17 Epstein, & Money, 1968; Hall et al., 2004; Hines, Brook, & Conway, 2004; Meyer-Bahlburg
18 et al., 2004; Nordenström, Servin, Bohlin, Larsson, & Wedell, 2002; Pasterski et al., 2005,
19 2007, 2011). There is also consistent evidence that women with CAH show reduced
20 heterosexual orientation. That is, they are more likely than women who do not have CAH to
21 report sexual attractions to, and encounters with, individuals of the same sex (Frisèn et al.,
22 2009; Hines, 2011; Meyer-Bahlburg, Dolezal, Baker, & New, 2008; Zucker et al., 1996).

23 In contrast to the consistent findings for alterations in gender role behavior and sexual
24 orientation, findings regarding gender identity in girls and women with CAH have been
25 inconsistent. *Gender identity* is a category of social identity and refers to the sense of self as

26 male, female, or a category that is neither male nor female. For the majority of the general
27 population, gender identity is congruent with biological parameters of sex (e.g.,
28 chromosomal, gonadal, and anatomic) and gender assignment at birth. However, some
29 individuals experience a psychological incongruence from their biological configuration
30 which manifests as distress around the intense and persistent wish to be the other sex
31 (American Psychiatric Association, 2013). According to the *Diagnostic and Statistical*
32 *Manual of Mental Disorders, Fourth Edition, Text Revision* (DSM-IV-TR) (American
33 Psychiatric Association, 2000), the relevant diagnosis for this condition for non-DSD
34 individuals was Gender Identity Disorder (GID) and in the new DSM-5 the relevant diagnosis
35 for individuals with and without DSD is Gender Dysphoria (GD) (American Psychiatric
36 Association, 2013). Although a change to live as the desired gender, along with hormone
37 therapy and surgery, is indicated as the most effective treatment for GID/GD that persists into
38 adulthood (Selvaggi & Bellringer, 2011), dysphoria¹ to this extent is rare; rates have been
39 approximated within the general population to be 0.005% to 0.014% for adult natal males and
40 0.002% to 0.003% for adult natal females (American Psychiatric Association, 2013).

41 With respect to gender dysphoria that has resulted in gender change in girls and
42 women with CAH, little is known about the developmental factors that may lead to such an
43 outcome. In a summary report of 250 individuals with CAH reared as girls, approximately
44 5.20% reported gender dysphoria and 30.0% of those dysphoric individuals changed to live
45 as men by adulthood (Dessens, Slijper, & Drop, 2005). While the rate of gender change from
46 female to male in 46,XX individuals with CAH is considerably higher than the rate in the

¹ Gender dysphoria here refers to unhappiness with the assigned gender, not to the DSM-5 diagnosis of Gender Dysphoria (GD). In this article, we distinguish these two concepts by using upper case for the initial letters of the two words of the DSM-5 diagnosis (Gender Dysphoria [GD]), and lower case for the initial letters of the words when used to describe unhappiness with the assigned gender (“gender dysphoria” or “dysphoria” in reference to a particular stated construct).

47 general population, alterations in gender identity have been less consistently observed than
48 alterations in gender role behavior or sexual orientation (Hines, 2011; Hines et al., 2004).

49 Analysis of individual studies assessing gender identification or gender dysphoria in
50 girls or women with CAH shows inconsistency of results. To date, 14 published studies have
51 reported gender identity outcomes in girls or women with CAH (see Table 1). Nine of the 14
52 studies suggest that girls or women with CAH have increased masculine gender identity
53 (Berenbaum & Bailey, 2003; Gupta et al., 2006; Hines et al., 2004; Hurtig & Rosenthal,
54 1987; Meyer-Bahlburg, Dolezal, Baker, Ehrhardt, & New, 2006; Slijper, 1984; Slijper, Drop,
55 Molenaar, & de Muinck Keizer-Schrama, 1998; Woelfle et al., 2012; Zucker et al., 1996),
56 whereas five of the 14 suggest that gender identity in girls or women with CAH does not
57 differ from that of control girls (Dittmann et al., 1990; Ehrhardt & Baker, 1974; Matilla,
58 Fagerholm, Santilla, Miettinen, & Taskinen, 2013; McGuire, Ryan, & Omenn, 1976; Meyer-
59 Bahlburg et al., 2004).

60 These inconsistent results may reflect methodological issues. First, there is a great
61 deal of inconsistency of measurement across studies. The only measure that has been used in
62 more than one study was the Draw-a-Person test (DAP), a projective measure that asks the
63 individual to draw a person, on the assumption that an individual generally draws a person
64 consistent with his or her own gender identity. Typically, people do draw a person of their
65 own gender but this is less likely to be the case among individuals diagnosed with GID
66 (Zucker, Finegan, Doering, & Bradley, 1983). The DAP was used in three studies of
67 individuals with CAH. Two studies of children found that girls with CAH were more likely
68 than other girls to draw a boy (Hurtig & Rosenthal, 1987; Slijper, 1984), whereas a study of
69 adults found no significant difference between women with and without CAH (McGuire et
70 al., 1975). None of the other studies used sufficiently similar methodology and age ranges to
71 allow direct comparison of results.

72 Second, several studies assessed gender identity using insufficient or unvalidated
73 measurement. For example, two assessments consisted of a single item (Ehrhardt & Baker,
74 1974) or a single item along with the DAP (McGuire et al., 1975; Slijper, 1984). One further
75 assessment consisted of only two items (Dittmann et al., 1990); another employed the DAP,
76 the Rorschach, and the Thematic Apperception Test (TAT), all of which are interpretive and
77 unvalidated for gender identity assessment (Hurtig & Rosenthal, 1987); and two studies
78 reported results based on physician impression (Gupta et al., 2006; Woefle et al., 2002).
79 These methods of assessment are difficult to interpret and could easily miss subtle differences
80 in gender identity of their participants.

81 Finally, of the 7 studies which employed diagnostic evaluations based on the DSM-IV
82 or scaled scores from self-report questionnaires/interviews, four confounded gender identity
83 with gender role behaviors, such as play or playmate preferences in childhood (Berenbaum &
84 Bailey, 2003; Meyer-Bahlburg et al., 2006; Slijper et al., 1998; Zucker et al., 1996). All four
85 of these studies reported increased cross-gender identification in girls or women with CAH,
86 but perhaps inflated by cross-gender role behavior. By contrast, of the three studies which
87 employed standardized assessment consisting purely of gender identity items, two reported
88 no effects in girls (Meyer-Bahlburg et al., 2004) or women (Mattila et al., 2013) with CAH.
89 However, the Mattila et al. study employed a measure which was developed for use with
90 adolescent and adult transsexuals and included questions about intentions for cross-sex
91 hormone therapy and sex reassignment surgery. Such a measure may not be sensitive enough
92 to detect more subtle alterations in gender identity. Hines et al. (2004) employed a six item
93 assessment pertaining only to gender identity and reported increased cross-gender
94 identification in 16 women with CAH.

95 There are other limitations to the existing studies, including small sample sizes, lack
96 of control groups, and wide age ranges of participants. Four of the existing studies did not

97 include controls for comparison purposes and most studies used small samples (range = 9 to
98 63 females with CAH, median = 18, see Table 1). Furthermore, age is an important variable
99 in relation to GID. Generally speaking, children who have been diagnosed with GID often no
100 longer meet the criteria when followed up in adolescence or adulthood (Green, 1987; Singh,
101 2012; Wallien & Cohen-Kettenis, 2008). If one were to apply this finding to females with
102 CAH, it would suggest that rates of GID may be higher in girls with CAH than in women
103 with CAH. Six studies included combined groups of girls and women with CAH. Three of
104 these reported increased cross-gender identification (Gupta et al., 2006; Slijper et al., 1998;
105 Woelfle et al., 2002), but the three others reported no increase (Dittmann et al., 1990; Matilla
106 et al., 2013; McGuire et al., 1975). For the three studies focusing only on women with CAH,
107 all three reported increased cross-gender identification (Hines et al., 2004; Meyer-Bahlburg et
108 al., 2006; Zucker et al., 1996), while only three of five studies focusing only on girls with
109 CAH reported this outcome (Berenbaum & Bailey, 2003; Hurtig & Rosenthal, 1987; Slijper,
110 1984).

111 Increased cross-gender identification in comparison to other girls or women does not
112 necessarily imply unhappiness with the assigned gender or gender dysphoria, so it is
113 important to consider evidence of possible gender dysphoria as well. While all but one
114 (Woelfle et al., 2002) of the reports listed in Table 1 presented data regarding a potential
115 increase in cross-gender identification, fewer gave rates of gender dysphoria. Where the
116 authors did not report these rates, relevant information was sometimes estimated from the
117 numbers of participants endorsing items suggesting possible gender dysphoria (e.g., “I am not
118 happy being a girl”) (Berenbaum & Bailey, 2003). Across all studies, 18/320 (5.62%) girls or
119 women with CAH were suspected by the authors to have experienced some degree of gender
120 dysphoria. In reports for girls only, 4/84 (4.76%) participants reported that they were not
121 happy being a girl, and for women only, 6/94 (6.38%) were considered to be dysphoric. Only

122 one of the 14 reports employed a clinical diagnostic interview (Slijper et al., 1998), however,
123 so these estimates cannot be assumed to equate to cases of GID or the DSM-5 diagnosis of
124 GD.

125 The convention in Western countries has been to assign 46,XX infants born with
126 classical CAH as female, given the potential for fertility, and to monitor development in
127 terms of psychosexual functioning where possible (Merke & Bornstein, 2005). For those who
128 are moderately to severely virilized, however, such an assignment may involve multiple
129 feminizing surgeries. In addition, the most severely virilized girls and women with CAH also
130 show the greatest degree of behavioral masculinization (Meyer-Bahlburg et al., 2008;
131 Nordenström et al., 2002). While alignment of gender identity, gender role behavior, and
132 sexual orientation as female-typical or male-typical is not essential for life satisfaction,
133 misalignment can be problematic. For example, gender identification contrary to assignment
134 may lead to psychological distress, or gender change later in life, involving additional surgery
135 and psychosocial challenges.

136 The aims of the current report were: (1) to provide empirical data concerning cross-
137 gender identification in girls with CAH using two standardized assessment measures and a
138 brief parent interview assessing behaviors related to cross-gender identification as
139 conceptualized in DSM-IV-TR; and (2) to investigate support for gender identity and gender
140 role behavior as distinct constructs using factor analysis. Boys with CAH were also studied
141 although no alterations were predicted in their gender identity.

142 **METHOD**

143 **Participants**

144 Parents of children with CAH were recruited through a national (United Kingdom)
145 CAH support group as well as through attending endocrinologists at 11 clinics throughout the
146 UK (representing England, Northern Ireland, Wales, and Scotland). Invitations through

147 endocrine clinics were made as general announcements of the study. Thirty-eight out of 95
148 invitations sent through the CAH support group were returned indicating acceptance for
149 participation, and resulted in the inclusion of 20 girls and 19 boys with CAH (one of the 38
150 invited families had two children with CAH). A further 23 girls and 19 boys with CAH
151 responded positively to invitations through the 11 endocrine clinics. Because there was a
152 great deal of overlap between members of the CAH support group and patient status at the 11
153 clinics, it was not possible to determine participation rates from clinics independent of the
154 support group. In addition, because recruitment was simultaneous at the 11 endocrine clinics
155 which were spread throughout a wide geographical area, it was not practical to identify exact
156 numbers of families who were or were not made aware of the study. In this case, determining
157 percentages of uptake of the general invitations made through endocrine clinics was not
158 possible. So that we could match participant children who had CAH to control children as
159 closely as possible, in terms of environmental characteristics (e.g., socioeconomic status,
160 parental education status, and family composition), we also invited siblings and first cousins
161 to participate.

162 Table 2 shows sample characteristics. Participants included 153 children: 43 girls with
163 classical CAH due to 21-hydroxylase deficiency (37 salt-wasting, 6 simple-virilizing); 38
164 boys with classical CAH due to 21-hydroxylase deficiency (35 salt-wasting, 3 simple-
165 virilizing); 41 female controls (31 sisters, 9 first cousins, 1 adopted sister); and 31 male
166 controls (23 brothers, 8 first cousins), all between 4- and 11-years of age. For each child, we
167 also interviewed a primary caregiver who was almost always the mother; six were fathers.
168 There were no significant group differences in age or in IQ, which was measured using the
169 Vocabulary subtest of the Wechsler Intelligence Scale for Children (4th ed.) (WISC-IV)
170 (Wechsler, 2003) or the Wechsler Preschool and Primary Scale of Intelligence (3rd ed.)
171 (WPPSI-III) (Wechsler, 2002) as age appropriate. Socioeconomic information and family

172 characteristics were obtained during an interview with parents. In our sample, 94.1% of child
173 participants were of Caucasian descent. The non-Caucasian participants were of Mid-Eastern
174 (1), Far Eastern (1), Indian/Bangladeshi (5), Afro-Caribbean (1), or other (1) descent. In our
175 sample, 96.1% of child participants came from two-parent families and 3.9% lived with a
176 mother only. With respect to parental level of education, 3.3% of the children came from
177 families where neither parent finished secondary school; 81.7% came from families where at
178 least one parent had vocational training/university degree; 13.7% came from families where
179 at least one parent had postgraduate education. Because we included unaffected siblings and
180 first cousins as controls, the demographic factors listed above were largely shared across
181 groups. Statistically there were no group differences in ethnic background, socioeconomic
182 status or family composition. All parent participants were fully informed with respect to
183 procedures and consented to their own and their child's inclusion in the study. Children who
184 were 7-years of age and older gave verbal assent. The protocol was approved by national and
185 institutional research ethics committees.

186 **Measures**

187 For a comprehensive assessment of gender identity, we administered two parent-
188 report measures (one interview and one questionnaire) and a child self-report interview as
189 follows.

190 *Gender Identity Interview for Children (GIIC)* (Wallien et al., 2009; Zucker et al., 1993)

191 This standardized measure, appropriate for children ages 2.5 to 12 years, contains 12
192 items assessing the child's cognitive and emotional understanding of his/her gender and/or
193 desire to be the other sex. Example questions are: "Are you a boy or a girl?" and "Do you
194 ever feel more like a boy than a girl?" Each item was scored by the interviewer as "0" for a

195 sex-appropriate response, as “1” for an “ambiguous” response, or as “2” for a “deviant²”
196 response, or cross-gender in relation to the participant’s assigned gender at birth. The total
197 score was the sum of item scores, ranging from 0 to 24. With respect to diagnostic usefulness
198 of the GIIC, employing a cut-off of 3+ or 4+ “cross gender” responses yielded high
199 specificity rates (88.8% and 93.9%, respectively) for GID in gender-referred children
200 (Wallien et al., 2009).

201 *Parent-report Gender Identity Questionnaire for Children (GIQC)* (Johnson et al., 2004)

202 The GIQC is a standardized measure developed to correspond to the core
203 phenomenology of the cross-gender identity and behaviors characteristic of a diagnosis of
204 GID, and is appropriate for children ages 2.5 to 12 years. Twelve items utilizing a 5-point
205 Likert response scale (e.g., 1 = every day to 5 = never) assessed gender-related play and
206 playmate preferences and a further 4 items assessed the desire to be the other sex and
207 anatomic dysphoria. With respect to diagnostic usefulness for GID, setting the specificity at
208 95% , the measure yielded a sensitivity rate of 86.8% for GID in gender-referred children
209 (Johnson et al., 2004). For the current analyses, the items pertaining to anatomic dysphoria
210 were excluded due to potential confound in participants with genital surgery or virilization.
211 *Parent Interview for Cross-gender Identification in Children (CIC)*

212 This parent interview was developed for the current study to tap the five behavioral
213 domains which comprise the cross-gender identification component (Part A) of GID as
214 described in the DSM-IV-TR. The aim was to assess whether girls with CAH engaged in
215 behaviors relevant to the diagnosis more so than unaffected girls. The format of the CIC was
216 devised to assess the frequency of these behaviors using a Likert scale. Specifically, the
217 behaviors assessed by the CIC included the following: (1) cross-gender identity statements,

² Note that “deviant” is the term applied by the authors of the measure to indicate cross-gender responses. For consistency, we use the original terminology.

218 (2) cross-dressing, (3) preference for cross-gender roles in role play, (4) preference for cross-
219 gender games and pastimes, and (5) preference for peers of the opposite gender. We used 3-
220 and 4-point response scales as follows: For items 1 and 2 (above), 1 = “frequently” to 4 =
221 “never;” and for items 3 to 5, 1 = “prefers cross-gender” 2 = “prefers neutral or equal cross-
222 gender/same-gender” and 3 = “prefers same-gender.” See Appendix A for all items in the
223 gender-specific versions of the CIC. For statistical analyses, we reversed these scores so that
224 high scores indicated increased cross-gender responses.

225 We did not assess Parts B, C, or D of the DSM-IV-TR criteria for GID for the
226 following reasons. Part B addresses “persistent discomfort with his or her sex” which is
227 assessed primarily in relation to genitalia. For girls, the criterion warrants “rejection of
228 urination in the sitting position, assertion that she has or will grow a penis, or assertion that
229 she does not want to grow breasts or menstruate.” Because many girls with CAH will have
230 been born with some degree of genital virilization, assessment of this criterion would
231 necessarily be confounded. Part C requires the exclusion of children diagnosed with “a
232 physical intersex condition.” Part D addresses clinically significant distress or impairment
233 and was not included because our assessment was part of a research study of gender
234 development in childhood and not a clinical evaluation.

235 **Statistical Analyses**

236 First, we analyzed the data for each of the three measures independently to assess
237 group effects and for comparison to prior published values where possible. We used 2 (Sex)
238 X 2 (CAH status) ANCOVA, with age as the covariate, to analyze the scale score data for the
239 GIIC and for the GIQC. For the CIC, we used Fisher’s exact test for group-wise comparisons.

240 Next, because gender identity is increasingly understood as a construct independent of
241 gender role behavior (Fagot, Leinbach, & Hagan, 1986; Meyer-Bahlburg et al., 2004; Zucker,
242 2010), we conducted analyses for composites of gender identity and gender role items

243 independently. To do this, we factor analyzed the items comprising the two measures in our
244 study which included both gender identity and gender role items, i.e., the GIQC and the CIC.
245 Specifically, we performed factor analysis using maximum likelihood as the extraction
246 method and a varimax rotated solution, across the entire sample of children ($N = 153$). Note
247 that in order to include male and female participants, the variables were structured to reflect
248 gender-congruent versus gender-incongruent (with respect to assigned gender), rather than
249 girl-typical or boy-typical, responses for each item. See Appendix B for factor loadings.

250 As we expected, the two identity items from the GIQC and the single identity item
251 from the CIC loaded on a single factor (Factor 2). The majority of the remaining items loaded
252 on two other factors. Factor 1 was populated by gender role items. Factor 3 included two
253 items (“plays with boy type dolls” and “imitates male characters”). Further inspection
254 showed that these two items were the least frequently endorsed of the male-typical activities,
255 as they were categorized in the original measure, for boys with and without CAH. This
256 suggests that these two items may not tap male-typical behavior as well as the other items do.
257 These two items were not included in further analyses for the composite variables. Finally,
258 we created standardized scores and calculated the mean for items from Factor 1 to create the
259 *gender role composite*, and repeated the process with items from Factor 2 to form the *gender*
260 *identity composite* (see Appendix B). ANCOVA with age as the covariate was used to
261 analyze the two composites.

262 RESULTS

263 Gender Identity Interview for Children (GIIC)

264 Table 3 shows the means (and SDs) for the GIIC. A 2 (Sex) x 2 (CAH status)
265 ANCOVA, with age covaried, revealed a significant main effect of age, $F(1, 152) = 6.81, p <$
266 $.05$, such that younger children gave more ambiguous/deviant responses, $r = -.21, p < .01$,
267 and a Sex X CAH status interaction, $F(1, 152) = 11.44, p < .01$. Simple effects analysis,

268 controlling for age, showed that girls with CAH gave significantly more ambiguous/deviant
 269 responses than any of the other groups: $F(1, 83) = 10.74, p < .01$, compared to control girls;
 270 $F(1, 80) = 19.20, p < .001$, compared to control boys; and $F(1, 73) = 9.39, p < .01$, compared
 271 to boys with CAH. There were no other significant group differences.

272 **Gender Identity Questionnaire for Children (GIQC)**

273 Table 3 shows means (and SDs) on the GIQC. A 2 (Sex) x 2 (CAH status) ANCOVA,
 274 with age covaried, revealed a significant Sex X CAH status interaction, $F(1, 152) = 47.54, p$
 275 $< .001$. Simple effects analysis indicated that, compared to control girls and boys with and
 276 without CAH, girls with CAH received significantly higher (increased cross-gender) scaled
 277 scores: $t(82) = 8.23, p < .001, t(79) = 9.94, p < .001, t(72) = -9.45, p < .001$, respectively.
 278 Furthermore, because this measure includes both gender identity and gender role behavior
 279 items, as demonstrated using factor analysis (described earlier), we calculated mean scores
 280 for these two subscales separately (see Factor Analysis for scale items). Two 2 (Sex) x 2
 281 (CAH status) ANCOVAs, with age as the covariate, for *gender identity* and *gender role*
 282 *behavior* revealed main effects of Sex [$F(1, 152) = 12.87, p < .001$ and $F(1, 152) = 9.03, p <$
 283 $.01$, respectively] and CAH status [$F(1, 152) = 139.29, p < .001$ and $F(1, 152) = 11.45, p <$
 284 $.001$, respectively] as well as Sex x CAH status interactions [$F(1, 152) = 12.50, p = .001$ and
 285 $F(1, 152) = 41.12, p < .001$, respectively]. There was no main effect of age in either ANCOVA.
 286 Group-wise comparisons revealed that girls with CAH received higher mean scores for
 287 *gender identity* items compared to controls girls and boys with and without CAH [$t(82) =$
 288 $3.82, p < .001, t(79) = 4.09, p < .001, t(72) = 3.90, p < .001$, respectively] as well as higher
 289 mean scores for *gender role behavior* items compared to controls girls and boys with and
 290 without CAH [$t(82) = 7.97, p < .001, t(79) = 12.48, p < .001, t(72) = 11.69, p < .001$,
 291 respectively].

292 **Parent Interview for Cross-gender Identification in Children (CIC)**

293 Table 4 shows the numbers (and %) of children who were reported by parents to have
294 displayed behavior in each of the five domains assessed by the CIC. Compared to children in
295 the other three groups (N = 105), significantly more girls with CAH stated the wish to be the
296 other gender (2/39 [5.1%] frequently stated the wish and 8/39 [20.5%] occasionally stated the
297 wish compared to 0/105 [0.0%] and 1/105 [1.0%]), respectively). Significantly more girls
298 with CAH were reported to frequently cross-dress: 12/39 (30.8%) compared to the other
299 children 1/105 (1.0%). Preferences for cross-gender roles in role play, for cross-gender
300 games, and peers of the opposite gender were also significantly more common among girls
301 with CAH (16/39 [41.0%], 20/39 [51.3%], and 17/39 [43.6%], respectively) compared to the
302 other children (2/105 [1.9%], 1/105 [1.0%], and 2/105 [1.9%], respectively). When we
303 considered the numbers of children in each group who exhibited behaviors in multiple
304 domains, we found that 10/39 (25.6%) of girls with CAH exhibited four out of the five and
305 5/39 (12.8%) exhibited all of the behaviors which comprise the cross-gender ideation
306 component of GID/GD,³ compared to 0/105 (0.0%) children in the other three groups in both
307 cases.

308 To test for statistically significant group differences, we used Fisher's exact test (due
309 to low expected values in some cells) to compare girls with CAH to control girls, control
310 boys, and boys with CAH with respect to frequency of displaying each of the five cross-
311 gender behaviors in question as well as for multiple cross-gender behaviors (e.g., 4/5
312 behaviors or 5/5 behavioral domains assessed). The results of these analyses are also shown
313 in Table 4. Girls with CAH showed increased cross-gendered responses compared to control

³ According to DSM-IV-TR (APA, 2000) diagnostic guidelines for GID, a combination of 4 out of the 5 behaviors in Part A (those behaviors addressed here in the CIC) must be present for a diagnosis. The latest manual, DSM-5 (APA, 2013), has implemented a stricter guideline whereby one of the 4 out of 5 exhibited behaviors must include the cross-gender identity statement, "wishes to be the other sex." In the current report, 5 out of 5 necessarily includes the cross-gender identity statement. We are not, however, claiming that these data are clinically diagnostic.

314 girls and boys with and without CAH in all five behavioral domains (range: $p < .001$ to $p <$
315 $.016$). In addition, girls with CAH were significantly more likely than children in any other
316 group to display behaviors simultaneously in 4/5 or 5/5 behavioral domains (ranges: $p < .001$
317 to $.002$, and $p < .029$ to $.048$, respectively).

318 **Gender Identity Composite and Gender Role Behavior Composite**

319 We conducted two 2 (Sex) X 2 (CAH status) ANCOVAs with age covaried for the
320 gender identity and gender role composites. Figure 1 shows the means and SE. For group
321 comparisons on the gender identity composite, there was a main effect of age, $F(1, 152) =$
322 $5.49, p < .05$, as well as a Sex X CAH status interaction, $F(1, 152) = 18.93, p < .001$.
323 Younger children showed more cross-gender identification, $r = -.19, p < .05$. Controlling for
324 age, girls with CAH showed more cross-gender identification as compared to all other
325 groups: control girls, $F(1, 83) = 18.54, p < .001, d = 1.13$; control boys, $F(1, 73) = 15.78, p <$
326 $.001, d = 1.19$; and boys with CAH, $F(1, 80) = 24.84, p < .001, d = 1.39$. There were no other
327 significant group differences.

328 On the gender role composite, there also was a significant Sex X CAH status
329 interaction, $F(1, 152) = 52.09, p < .001$. However, in this case, age was not significant. Girls
330 with CAH showed more cross-gender role behavior as compared to all other groups: control
331 girls, $t(82) = 9.17, p < .001, d = 1.99$; control boys, $t(72) = 11.26, p < .001, d = 2.80$; and
332 boys with CAH, $t(79) = 12.19, p < .001, d = 2.59$. There were no other significant group
333 differences.

334 Because gender identity and gender role behavior typically co-vary, we conducted
335 bivariate correlations for the two composite scores. For the whole sample, the correlation was
336 $r(151) = .56, p < .001$. Within groups, the correlations were: $r(41) = .42, p < .01$ for girls
337 with CAH; $r(39) = .19, ns$ for control girls; and $r(29) = .193, ns$ for control boys. A

338 correlation coefficient could not be calculated for boys with CAH as there was no variance
339 (i.e., $SD = 0.0$) in scores on the gender identity composite for this group.

340 **DISCUSSION**

341 The current report makes three important contributions to understanding the
342 relationship between prenatal androgen exposure and gender identity. First, it provides robust
343 evidence, using dimensional self- and parent-report measures of cross-gender identification
344 that girls exposed to high levels of androgens prenatally because they have CAH show
345 increased cross-gender identification, independent of gender role behavior, compared to
346 female controls. Unlike prior studies, we used interview and questionnaire methods with
347 parents, as well as direct assessment with children, and found consistent evidence across all
348 three measures. Second, using factor analysis we found clear support for the distinctness of
349 the two constructs, gender identity and gender role, which have often been conflated in prior
350 studies. Finally, our findings have implications for understanding the role of prenatal
351 androgen exposure more generally in the development of gender identity by showing that
352 degree of gender identification congruent with biological (chromosomal, gonadal, and
353 hormonal) manifestations of gender, at least in girls, are sensitive to androgen exposure *in*
354 *utero*, in a similar manner to gender role behavior and sexual orientation.

355 Compared to girls without CAH and boys with and without CAH, girls with CAH in
356 our sample gave self-reports and were reported by their parents to exhibit greater cross-
357 gender identification as well as greater cross-gender role behavior. Using a child-report
358 interview schedule, the GIIC, we found that girls with CAH gave significantly more
359 “ambiguous” or “deviant” responses to questions such as, “Do you ever feel more like a boy
360 than a girl?” or “Do you ever think you really are a boy?” Furthermore, parents reported
361 similar patterns of behavior in questionnaire and interview measures. On the GIQC, parents
362 of girls with CAH reported that their daughters showed greater cross-gender identification as

363 well as cross-gender role behavior compared to parents of children in the other three groups.
364 Lastly, we administered a parent interview developed for the current study and aimed at
365 assessing the frequency of engagement in a subset of behaviors characteristic of GID as
366 conceptualized in DSM IV-TR and DSM5. We found that compared to percentages of
367 children in the other three groups (girls without CAH and boys with and without CAH),
368 greater numbers of girls with CAH were reported by their parents to have engaged in the
369 behaviors in question (e.g., making cross-gender statements, cross-dressing, taking cross-
370 gender roles in make-believe play) with frequency consistent with GID/GD.

371 Findings from assessments in our study can also be compared to those of children in
372 the general population who have been referred to gender clinics because of cross-gender
373 behavior, since those children have also been assessed using the GIIC and the GIQC. On the
374 GIIC (Wallien et al., 2009), samples of children referred to gender clinics and who were
375 diagnosed with GID, obtained higher mean item scores for cross-gender identification
376 compared to girls with CAH in the current study, but the children referred to gender clinics
377 who were subthreshold for the GID diagnosis and control children obtained lower mean item
378 scores for cross-gender identification than these girls with CAH. Compared to previously
379 reported data on the GIQC (Johnson et al., 2004), girls with CAH in our study showed lower
380 mean item scores for cross-gender identification and cross-gender role behavior than girls
381 referred to gender clinics, who were diagnosed with GID, but showed similar mean item
382 scores to children referred to gender clinics who were subthreshold for the GID diagnosis and
383 higher mean item scores than control children. These results suggest that, on average, girls
384 with CAH show at least as much cross-gender identification as girls without CAH who have
385 problems sufficient to be referred to a gender clinic.

386 Finally, our findings lend support to the distinctness of gender identity and gender
387 role behavior. Using factor analysis, we found that items asking directly about the wish to be

388 the other gender loaded together on a factor separate from gender role behavior. Analyzing
389 the two new composite variables, the gender identity composite and the gender role behavior
390 composite, we again found that, compared to girls without CAH and boys with and without
391 CAH, significantly more girls with CAH stated the desire to be the other gender and they
392 exhibited more cross-gender role behaviors. Although the gender identity and gender role
393 behavior composite scores were shifted in the masculine direction in girls with CAH, the two
394 constructs showed a different relationship with age. Age negatively covaried with gender
395 identity but not gender role behavior. Furthermore, while scores for the two composites were
396 significantly correlated within the sample as a whole [$r(151) = .56, p < .001$], closer
397 inspection showed that the correlation was driven by scores for the group of girls with CAH
398 [$r(41) = .42, p < .01$]. This correlation was not significant for any of the other three groups,
399 primarily due to a lack of variance in the gender identity composite, e.g., $SD = 0.00$ for the
400 mean gender identity composite scores for boys with CAH. The differing patterns of
401 covariance for the two composites with age, as well as the lack of correlation between the
402 two composites for children other than girls with CAH, lend further support to the need to
403 consider gender identity and gender role behavior as two separate constructs.

404 With respect to limitations of the current report, it is possible that girls with CAH who
405 have experienced gender identity difficulties were over- or under-represented as a result of
406 selection bias. For instance, it is possible that parents of girls who showed signs of cross-
407 gender identification were more likely to participate in the study. Alternatively, the bias could
408 be that girls who were *less* cross-gendered in their identity were more likely to participate.
409 The possibility of selection bias is unavoidable when studying rare and potentially
410 stigmatizing disorders, given that participation is voluntary (Pasterski, Mastroyannopoulou,
411 Wright, & Hughes, 2013). Prior studies of gender identity and other gender-related behaviors
412 in individuals with CAH would have been subject to similar biases. Importantly, our findings

413 for childhood gender role behavior appear to be similar to those reported in prior studies. For
414 example, Meyer-Bahlburg et al. (2004) reported an effect size of $d = 1.75$ between girls with
415 and without CAH for gender role behavior compared to $d = 1.99$ for the current study. By
416 contrast, however, the same study (Meyer-Bahlburg et al., 2004) reported an effect size of $d =$
417 -0.16 for gender identity between girls with and without CAH using the GIIC (Wallien et al.,
418 2009; Zucker et al., 1993) whereas we found $d = 0.80$ for the same measure, and $d = 1.11$ on
419 our gender identity composite.

420 It is possible that cohort effects contributed to our observing higher rates of cross-
421 gender identification than seen in prior studies. Recent reports suggest that the rate of referral
422 for gender dysphoria in the general population is increasing for both males and females
423 (Zucker, Wood, & VanderLaan, 2014). One of the more plausible explanations for this
424 increase may be that gender variant behavior has become more widely accepted and seeking
425 social support for distress related to gender dysphoria may be less stigmatized than in the
426 past. It is possible that changes in social acceptance of gender variant behavior might have
427 contributed to differences between the results of the current study and those of prior studies.

428 **Clinical Implications**

429 Information about the determinants of gender identity is important for making
430 decisions about gender assignment, not only in girls with CAH, but in cases of genital
431 ambiguity in other DSD, where there is an incongruence of chromosomal, hormonal, and
432 anatomic sex. Many DSD involve exposure to atypical concentrations of androgenic
433 hormones and this atypical exposure might be expected to influence gender identification.
434 Based on the current findings, it might also be expected to increase the likelihood of cross-
435 gender identification.

436 With respect to CAH specifically, girls with CAH in the current study showed
437 significantly increased cross-gender identification and cross-gender role behavior using a

438 comprehensive and multi-method assessment. In adulthood, some women with CAH have
439 either transitioned to living in the male gender role or have seriously considered such a
440 transition (Dessens et al., 2005; Meyer-Bahlburg, 1996; Meyer-Bahlburg et al., 2006). Our
441 findings suggest that younger females with CAH may already be questioning their gender
442 identity.

443 It is important also to consider that many children who have been diagnosed with GID
444 have been found to desist in their desire to change gender as they get older (de Vries &
445 Cohen-Kettenis, 2012; Wallien & Cohen-Kettenis, 2008). In light of this desistance, it would
446 be of interest to conduct comprehensive, longitudinal assessments of gender identity and
447 gender dysphoria in substantial samples of girls/women with CAH. This would provide some
448 evidence as to whether girls with CAH who show evidence of gender dysphoria are similarly
449 likely to desist prior to adulthood, as are other girls. Our findings that age negatively
450 correlated with the GIIC score and with scores for the gender identity composite suggest that
451 some percentage of girls with CAH presenting with elements of cross-gender identification
452 may also desist. Unfortunately, however, we cannot extrapolate longitudinal information
453 from cross-sectional data. Studies of women with CAH indicate similar, rather than reduced,
454 rates of cross-gender identification compared to those seen in girls with CAH. For example,
455 considering rates of gender dysphoria reported in previous studies combined, a total of 4/84
456 (4.7%) girls with CAH showed some signs of gender dysphoria (Berenbaum & Bailey, 2003;
457 Ehrhardt & Baker, 1974, Hurtig & Rosenthal, 1987; Meyer-Bahlburg et al., 2004) whereas, in
458 studies of adults, a total of 5/110 (4.5%) women with CAH showed some signs of gender
459 dysphoria (Hines et al., 2004; Meyer-Bahlburg et al., 2006; Zucker et al., 1996), and among
460 studies including groups of children/adolescents/adults combined, the comparable numbers
461 were 8/142 (5.63%) of participants (Dittmann et al., 1990; Gupta et al., 2006; Matilla et al.,
462 2013; McGuire et al., 1975; Slijper et al., 1998; Woelfle et al., 2002). Again, although these

463 calculations might seem to suggest that the level of gender dysphoria among girls with CAH
464 is similar to that seen among women with CAH, longitudinal data are needed to address the
465 question of desistance rigorously.

466 Though physical gender change from female to male in cases of 46,XX women with
467 CAH is relatively uncommon (~1.6%) (Dessens et al., 2005), it occurs at a greater frequency
468 than in the general population (~.003%) (American Psychiatric Association, 2013). The
469 developmental factors that led to gender change in these individuals with 46,XX CAH are
470 unknown. It is also unclear whether shifts toward cross-gender identification cause distress in
471 girls with CAH. It is possible that, while some girls and women with CAH may wish to be
472 the other gender, some may have found coping strategies which alleviate potential distress.
473 Future studies could usefully assess the psychological and emotional implications of cross-
474 gender identification in girls with CAH.

475 Finally, with respect to gender assignment (or reassignment), cross-gender
476 identification in girls with CAH is relevant to decision making, though long-term gender
477 identity is not the sole factor in judging a successful assignment. Studies reporting on
478 satisfaction with cosmetic genitoplasty suggest that women who have had many surgeries,
479 due to severe virilization, have poorer psychosocial/psychosexual outcomes than those with
480 less virilization and fewer surgeries (Nordenström et al., 2010). Furthermore, these poorer
481 surgical outcomes correlated with poorer quality of life on factors such as sexual functioning
482 and satisfying romantic relationships (Nordenskjöld et al., 2008; Nordenström et al., 2010). If
483 those with the greatest virilization also show the greatest levels of cross-gender identification,
484 as they do in regard to gender role behavior and sexual orientation (Hines, 2011), perhaps
485 greater overall life satisfaction may be found with male sex-assignment for some of the most
486 strongly virilized girls with CAH (Houk & Lee, 2010; Lee, Houk, & Husmann, 2010). In
487 considering male sex assignment for such individuals, however, it is important to consider the

488 lack of fertility that such assignment would entail, although some research suggests that lack
489 of fertility is of less concern to severely virilized females with CAH than it is to other women
490 (Casteras, De Silva, & Rumsby, 2009). In addition, recent advances in reproductive
491 technologies could ameliorate the negative aspects of the inability to conceive without
492 assistance. Future studies addressing the relationship between virilization and gender identity
493 could further inform policy regarding gender assignment in severely virilized 46,XX CAH.

494 **Summary**

495 The current report provides robust and comprehensive evidence that girls with CAH
496 show increased cross-gender identification compared to controls and to boys with CAH.
497 Given that cross-gender identification and cross-gender role behavior are characteristic of
498 children with GID/GD, and considering the frequency of stated cross-gender identification
499 and engagement in cross-gender role behaviors reported by parents of girls with CAH, our
500 findings suggest that the potential for gender dysphoria in girls with CAH may be higher than
501 prior reports have suggested. Future studies may clarify the link between cross-gender
502 identification and gender dysphoria by explicitly assessing levels of distress and/or
503 impairment which is also a key feature of GID/GD. Furthermore, we showed gender identity
504 and gender role behavior to be distinct constructs, using factor analysis of scale items
505 typically included together in gender identity assessments. This suggests that some previous
506 reports on gender identity may have confounded gender identity and gender role in their
507 assessments. Finally, we have provided information relevant to the role of prenatal androgen
508 in the development of gender identity more generally. Our findings suggest that prenatal
509 androgen exposure, at least in girls, is related to increased cross-gender identification as well
510 as increased cross-gender role behavior.

Appendix A

Parent Interview for Cross-gender Identification in Children (CIC)

Girl Form*

1. *Identity statements*

- Child frequently states the wish to be of the opposite sex or that she is a member of the opposite sex (e.g., “I want to be a boy, I am a boy, I want to grow up to be a daddy, not a mommy”).
- Child occasionally states the wish to be of the opposite sex.
- Child rarely states the wish to be of the opposite sex.
- Child does not state the wish to be of the opposite sex.

2. *Cross-dressing*

- Child frequently cross-dresses (e.g., refuses to wear culturally typical feminine clothing, and prefers boys’ trousers and shirts to simulate a masculine appearance; desires hair to be cut very short).
- Child occasionally cross-dresses.
- Child rarely cross-dresses.
- Child never cross-dresses.

3. *Role play*

- Child prefers to engage in roles that are traditionally associated with the opposite sex (e.g., a preference for role play as a male; emulation of male superheroes; drawings are invariably of men).
- Child engages in roles that are both traditionally masculine and feminine.
- Child avoids toys and roles that are traditionally feminine but does not engage in toy or role play that is traditionally masculine (i.e., prefers neutral activities such as drawing or making music).
- Child prefers to engage in roles that are traditionally feminine.

4. *Games and pastimes*

- Child prefers to play games / engage in pastimes that are traditionally masculine (e.g., rough-and-tumble play, wrestling, sports with boys).
- Child plays games / engages in pastimes that are both traditionally masculine and feminine.
- Child avoids games / pastimes that are traditionally feminine but does not play games / engage in pastimes that are traditionally masculine (i.e., prefers neutral games and pastimes).
- Child prefers to play games / engage in pastimes that are traditionally feminine.

5. *Peer relations*

- Child prefers to play with opposite-sex peers.
- Child is a loner or is rejected.
- Child plays with both same-sex and opposite-sex peers.
- Child prefers to play with same-sex peers.

* The Boy Form for this measure was identical, with relevant gendered terms reversed to reflect same or cross-sex preferences/behaviors for a boy.

Appendix B

Factor loadings for the Parent Interview for Cross-gender Identification in Children (CIC) and the Parent report Gender Identity Questionnaire for Children (GIQC).

Measure	Item	Factor loadings*		
		1	2	3
CIC	Cross-dressing	.58	.39	.07
CIC	Role play	.87	.18	.04
CIC	Peer relations (same or other sex)	.61	.35	.12
CIC	Games and pastimes	.83	.14	.03
GIQC	Playmates (same or other sex)	.64	.26	.14
GIQC	Plays with Barbie	.78	.11	-.45
GIQC	Plays with make-up	.75	.19	-.36
GIQC	Imitates female characters	.65	.25	-.52
GIQC	Plays sports with boys (but not girls)	.60	.18	-.03
GIQC	Plays same sex in playing house	.71	.23	.04
GIQC	Plays girl-type games (compared to boy-type)	.68	.16	-.20
GIQC	Plays boy-type games (compared to girl-type)	.72	.15	.05
GIQC	Dresses up in same or other sex clothes	.83	.26	.02
CIC	States that s/he is the other sex	.30	.89	.04
GIQC	States wish to be other sex	.25	.80	.06
GIQC	States that s/he is the other sex	.25	.59	-.08
GIQC	Plays with GI Joe	.00	-.09	.83
GIQC	Imitates male characters	.42	.09	.61
GIQC	Plays sports with girls (but not boys)	-.10	.18	.26

*Factor 1 items constitute the gender role composite; Factor 2 items constitute the gender identity composite. Factor 3 items appear to be less consistent with gender role or gender identity compared to other items. Further inspection showed these three items to be rated as the least frequent of the male-typical activities among boys, suggesting that they may not discriminate male-typical behavior as well as the other items do. These two items were not included in further analyses for the composite variables.

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Table 1. Studies investigating gender identity in girls and women with congenital adrenal hyperplasia (CAH).

	Child/Adolescent (< 19 years)	Adults (>18years)	Child Adolescent & Adult	Measurement Of Gender Identity/Dysphoria	Evidence of Increased Cross-Gender Identification ^a	Gender Dysphoria
Ehrhardt & Baker (1974)	17 CAH 11 controls			Single item self-report	No	0/17 (0%) ^b
McGuire et al. (1975)			15 CAH 15 Controls	Single item self-report & Draw-A-Person test ^c	No	0/15 (0%)
Slijper (1984)	25 CAH 97 controls			Single item self-report & Draw-A-Person test	Yes	Not enough information
Hurtig and Rosenthal (1987)	9 CAH 6 Controls			Draw-A-Person test, Rorschach, & Thematic Apperception Test	Yes	0/9 (0%)
Dittmann et al. (1990)			35 CAH 16 controls	Two item self-report assessment	No	0/35 (0%)
Zucker et al. (1996)		31 CAH 15 controls		Standardized assessment (self-report, recalled)	Yes	0/31 (0%) ^d
Slijper et al. (1998)			18 CAH	Diagnostic interview DSM-IV	Yes	2/18 (11%)
Woelfle et al. (2002) ^e			9 CAH	Physician impressions	---	1/9 (11%)
Berenbaum and Bailey (2003)	43 CAH 36 controls			Standardized interview (self-report)	Yes	4/43 (9%) ^f
Hines et al. (2004)		16 CAH 15 controls		Six item assessment (self-report, recalled)	Yes	Not enough information
Meyer-Bahlburg et al. (2004)	15 CAH 30 controls			Standardized assessment (self-report)	No	0/15 (0%)
Meyer-Bahlburg et al. (2006)		63 CAH 24 controls		Standardized assessment (self-report)	Yes	5/63 (8%)
Gupta et al. (2006)			50 CAH	Physician interview	Yes	5/50 (10%)
Matilla et al. (2013)			15 CAH	Standardized assessment (self-report)	No	0/15 (0%)

^aConclusion drawn from data presented in each study. An effect was considered to be present if scores were significantly shifted in comparison to controls or normative data.

^bNote that 37% of girls with CAH compared to <1% of unaffected sisters reported that they would have preferred to have been a boy if the choice had been possible.

^cDraw-a-Person test (DAP). The first figure drawn is meant to indicate gender identification with males or females.

^dOne proband who refused participation was diagnosed as transsexual. Inclusion of this proband produces an incidence of 3.2% gender dysphoria in the CAH group.

^e3/9 patients were reassigned female between ages 7 and 9 years including the one reported to be gender dysphoric (reassigned to female at 7 years old).

^fAs indicated by endorsement of the item stating that she is not happy being a girl; None of the controls endorsed this item.

Table 2. Sample characteristics.

	Girls		Boys	
	CAH	Control	CAH	Control
	N = 43	N = 41	N = 38	N = 31
Age (years) ^a	<hr/>			
M	7.13	7.59	7.15	7.81
SD	2.28	2.51	2.04	2.36
Range	4.01-11.90	4.00-11.86	4.06-11.34	4.17-11.93
IQ Vocab subtest ^b	<hr/>			
M	10.02	11.12	10.74	10.84
SD	3.33	2.99	2.71	2.56
Parent informant ^c	<hr/>			
Mothers	41	40	38	29
Fathers	2	1	1	2

^aThere were no significant group differences in age.

^bThe Vocabulary subtest was used as a proxy of Full-scale IQ using the WPPSI (Wechsler, 1967, 2002) and WISC (Wechsler, 2003) as age appropriate. There were no significant group differences on this measure.

^cNo analyses were carried out comparing mothers and fathers given the low number of fathers participating. Parent-report data reflect the distribution listed here.

Table 3. Means (SDs) and group comparisons for the *Gender Identity Interview for Children* (GIIC) and the *Gender Identity Questionnaire for Children* (GIQC; parent-report).

Group	M	SD	N	Groups compared to CAH girls	
Gender Identity Interview for Children (GIIC; high = cross-gender)					
				<i>p</i>	<i>d</i> [†]
CAH girls	3.80	3.50	43	---	---
Control girls	1.59	2.00	41	.001	0.82
CAH boys	1.00	1.58	31	<.001	1.14
Control boys	1.37	1.90	38	<.001	1.00
Gender Identity Questionnaire for Children (GIQC; high = cross-gender)					
GIQC Total Score					
				<i>p</i>	<i>d</i> [†]
CAH girls	2.96	0.63	43	---	---
Control girls	2.02	0.40	41	<.001	1.85
CAH boys	1.92	0.25	38	<.001	2.44
Control boys	1.95	0.27	31	<.001	2.44
GIQC Gender Identification					
				<i>p</i>	<i>d</i> [†]
CAH girls	1.44	0.71	43	---	---
Control girls	1.02	0.11	41	<.001	1.03
CAH boys	1.00	0.00	38	<.001	1.24
Control boys	1.02	0.09	31	<.001	1.08
GIQC Gender Role Behavior					
				<i>p</i>	<i>d</i> [†]
CAH girls	3.34	0.77	43	---	---
Control girls	2.20	0.51	41	<.001	1.81
CAH boys	1.69	0.36	38	<.001	3.03
Control boys	1.71	0.41	31	<.001	3.06

[†]Effect sizes are Cohen's *d* (Cohen, 1988).

Table 4. Numbers (and %) of children in each group reported to engage in behaviors characteristic of cross-gender identification assessed using the Parent Interview for Cross-gender Identification for Children (CIC).

	Girls		Boys		<i>(p-values for Fisher's Exact Test)</i>			
	CAH N = 39	Control N = 38	CAH N = 36	Control N = 31	Girls with CAH compared to: Control Girls Boys with CAH Control Boys			
1. Wishes to be the other sex	(Frequently)	2 (5.1%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	<i>ns</i>	<i>ns</i>	<i>ns</i>
	(Occasionally)	8 (20.5%)	1 (2.6%)	0 (0.0%)	0 (0.0%)	.016	.004	.008
2. Cross-dresses	(Frequently)	12 (30.8%)	0 (0.0%)	0 (0.0%)	1 (3.2%)	< .001	< .001	.003
	(Occasionally)	3 (7.7%)	1 (2.6%)	3 (8.3%)	1 (3.2%)	<i>ns</i>	<i>ns</i>	<i>ns</i>
3. Prefers opposite-sex roles only		16 (41.0%)	1 (2.6%)	0 (0.0%)	1 (3.2%)	< .001	< .001	< .001
4. Prefers opposite-sex games only		20 (51.3%)	0 (0.0%)	1 (2.8%)	0 (0.0%)	< .001	< .001	< .001
5. Prefers opposite-sex peers only		17 (43.6%)	2 (5.2%)	0 (0.0%)	0 (0.0%)	< .001	< .001	< .001
• Engages in 4/5 of the behaviors listed above[†]		10 (25.6%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	.001	.001	.002
• Engages in 5/5 of the behaviors listed above[†]		5 (12.8%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	.029	.033	.048

[†]Includes “frequently” or “occasionally” for “wishes to be the other sex” and “cross dresses.”

Table 4. Means (SDs) and group comparisons for the *Gender Identity Interview for Children* (GIIC) and the *Gender Identity Questionnaire for Children* (GIQC; parent-report).

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[†]Effect sizes are Cohen's *d* (Cohen, 1988).

Figure 1. Z-scores for gender role behavior and gender identification composites. High scores are cross-gender; bars represent SE; Group comparisons controlled for age.

*** $p < .001$ for comparisons of girls with CAH to each of the three other groups.

Figure 1.

