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

INTRODUCTION

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

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

male, female, or a category that is neither male nor female. For the majority of the general population, gender identity is congruent with biological parameters of sex (e.g., chromosomal, gonadal, and anatomic) and gender assignment at birth. However, some individuals experience a psychological incongruence from their biological configuration which manifests as distress around the intense and persistent wish to be the other sex (American Psychiatric Association, 2013). According to the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision* (DSM-IV-TR) (American Psychiatric Association, 2000), the relevant diagnosis for this condition for non-DSD individuals was Gender Identity Disorder (GID) and in the new DSM-5 the relevant diagnosis for individuals with and without DSD is Gender Dysphoria (GD) (American Psychiatric Association, 2013). Although a change to live as the desired gender, along with hormone therapy and surgery, is indicated as the most effective treatment for GID/GD that persists into adulthood (Selvaggi & Bellringer, 2011), dysphoria[1] to this extent is rare; rates have been approximated within the general population to be 0.005% to 0.014% for adult natal males and 0.002% to 0.003% for adult natal females (American Psychiatric Association, 2013).

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

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

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

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

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

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

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

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

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

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

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

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

METHOD

Participants

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

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

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

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

Measures

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

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

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

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

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

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

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

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

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

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

Statistical Analyses

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

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

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

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

RESULTS

Gender Identity Interview for Children (GIIC)

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

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

Gender Identity Questionnaire for Children (GIQC)

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

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

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

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

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

Gender Identity Composite and Gender Role Behavior Composite

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

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

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

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

DISCUSSION

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

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

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

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

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

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

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

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

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

Clinical Implications

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

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

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

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

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

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

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

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

Summary

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

[a]Conclusion 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.
[b]Note 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.
[c]Draw-a-Person test (DAP). The first figure drawn is meant to indicate gender identification with males or females.
[d]One proband who refused participation was diagnosed as transsexual. Inclusion of this proband produces an incidence of 3.2% gender dysphoria in the CAH group.
[e]3/9 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).
[f]As 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]				
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]				
M	10.02	11.12	10.74	10.84
SD	3.33	2.99	2.71	2.56
Parent informant[c]				
Mothers	41	40	38	29
Fathers	2	1	1	2

[a] There were no significant group differences in age.
[b] The 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.
[c] No 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)				*p*	*d*†
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				*p*	*d*†
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				*p*	*d*†
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				*p*	*d*†
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		(*p*-values for Fisher's Exact Test) Girls with CAH compared to:		
	CAH N = 39	**Control N = 38**	**CAH N = 36**	**Control N = 31**	**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%)	*ns*	*ns*	*ns*
(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%)	*ns*	*ns*	*ns*
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).

Group	M	SD	N	Groups compared to CAH girls	
Gender Identity Interview for Children (GIIC; high = cross-gender)				*p*	*d*†
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				*p*	*d*†
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Control girls	2.02	0.40	41	< .001	1.85
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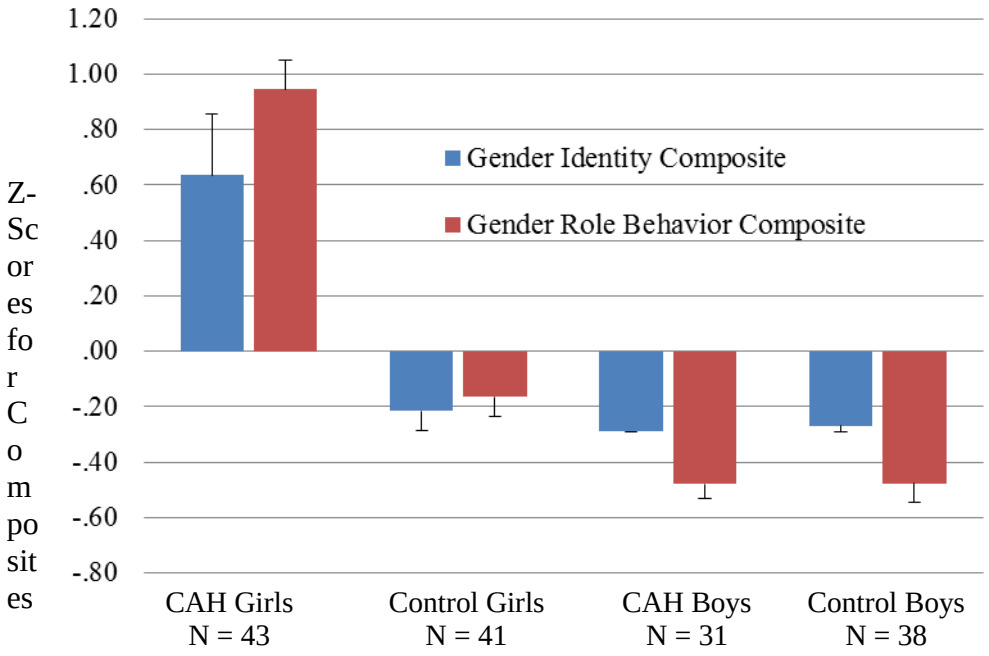
Control boys	1.95	0.27	31	< .001	2.44
GIQC Gender Identification				*p*	*d*†
CAH girls	1.44	0.71	43	---	---
Control girls	1.02	0.11	41	< .001	1.03
CAH boys	1.00	0.00	38	< .001	1.24
Control boys	1.02	0.09	31	< .001	1.08
GIQC Gender Role Behavior				*p*	*d*†
CAH girls	3.34	0.77	43	---	---
Control girls	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).

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.


1.20
1.00
.80
.60
.40
.20
.00
-.20
-.40
-.60
-.80
Z-Scores for Composites
Gender Identity Composite
Gender Role Behavior Composite
CAH Girls N = 43
Control Girls N = 41
CAH Boys N = 31
Control Boys N = 38