

Effects on Gender Identity of Prenatal Androgens and Genital Appearance: Evidence from Girls with Congenital Adrenal Hyperplasia

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To address questions about sex assignment in children with ambiguous genitalia, we studied gender identity in girls with congenital adrenal hyperplasia (CAH) in relation to characteristics of the disease and treatment, particularly genital appearance and surgery. A 9-item gender identity interview was administered to 43 girls with classical CAH ranging in age from 3–18 yr, 7 tomboys, and 29 sister control girls. Groups were compared on total score and on individual items. Results showed that, on the total gender identity score, 88% of girls with CAH had scores overlapping those of control girls, but

the average score was intermediate between control girls and tomboys. On individual items of gender identity (discomfort as a girl, wish to be a boy), girls with CAH were similar to control girls. Gender identity in girls with CAH was not related to degree of genital virilization or age at which genital reconstructive surgery was done. Thus, moderate androgen excess early in development appears to produce a small increase in the risk of atypical gender identity, but this risk cannot be predicted from genital virilization. (*J Clin Endocrinol Metab* 88: 1102–1106, 2003)

RECENT CONTROVERSIES HAVE renewed interest in the determinants of gender identity. For almost 50 yr, treatment of individuals with intersex conditions was shaped by the belief that gender identity results from social rearing rather than biological factors (1–4). Infants with ambiguous genitalia and infant males with a severely malformed or damaged penis have generally been assigned to the female sex because it is easier to construct female rather than male genitalia and because female gender identity was assumed to result from female-typical socialization, provided that gender-confirming genital surgery is done early in life.

This practice and the assumptions underlying it have been challenged in two ways. The first is a reassertion of the primacy of prenatal hormones in determining gender identity. This position has been supported by a case report of an individual reared as a girl after a mishandled circumcision but who experienced gender dysphoria and requested sex reassignment (5, 6), and by a report that many XY males with absent or malformed penis due to cloacal exstrophy who were reared as females declare themselves to be boys (7). But other evidence suggests caution in concluding that gender identity is determined primarily by prenatal androgen exposure. The publicized individual with ablatio penis (6) was reared as a boy early in life; another individual with a similar history but with earlier female reassignment had a different outcome, particularly female gender identity (8). To date, there have been no published systematic studies of individuals with cloacal exstrophy. Case reports and small-scale studies of individuals with cloacal exstrophy and other intersex conditions, including micropenis and partial androgen insensitivity syndrome, suggest variations in gender

identity outcome that are not predictable from biological or social factors (7, 9–11).

The second controversial aspect of current medical treatment for children with ambiguous genitalia concerns the practice of genital reconstructive surgery early in life, especially given its potential risk in impairing adult sexual response. Gender identity development and psychological adjustment are assumed to require concordance between genital appearance and rearing sex (3), but this assumption has not been systematically tested. Challenges to this practice have resulted in discussions about deferring surgery in intersex individuals (12–14), although there is little evidence concerning the psychological benefits and costs of early *vs.* late surgery. Case reports suggest that gender identity can develop in the absence of sex-typical external genitalia (as in cloacal exstrophy and micropenis; Refs. 6, 7, 15, and 16). Furthermore, gender dysphoria occurs in individuals with sex-typical genitalia and no known disorder of physical sexual differentiation (17). Sex-typical genitalia thus appear to be neither necessary nor sufficient for sex-typical gender identity, but may be related in a way not yet specified.

In light of the current controversies about the medical management of children with intersex conditions and the impact of medical decisions on the patient's well-being, it becomes increasingly important to understand the determinants of gender identity. This question can be effectively studied in females with congenital adrenal hyperplasia due to 21-hydroxylase deficiency (CAH) because the disorder is relatively common, there is dissociation of prenatal androgen exposure and rearing sex, and variation in gender identity can be examined in relation to variations in genital appearance and genital surgery.

There is much less systematic data on gender identity in females with CAH than there is on other aspects of sex-

Abbreviation: CAH, Congenital adrenal hyperplasia.

related behavior. In the only study of gender identity in young females with CAH, none of the 17 subjects had a conflict with her female gender identity or was unhappy being a girl, but girls with CAH were more likely than controls to think that it might be better to be a boy (18). Gender identity was not systematically assessed, so it is difficult to know whether the latter reflects a manifestation of interest in boy-typical play (19). Studies of adult females with CAH indicate that a small minority identify as males, higher than the population rate of transsexualism (20–22). In the only systematic study of gender identity, adult women with CAH were not gender dysphoric but did differ from unaffected sisters in degree of gender identity, reporting less childhood and adult sense of femininity (22). Simple-virilizers had more atypical identification than did salt-wasters, suggesting that degree of prenatal androgen exposure is not a key determinant of gender identity, given that salt-wasting CAH is associated with higher levels of prenatal androgens than simple-virilizing CAH (23, 24). This study illustrates the importance of using systematic assessments of gender identity but is limited by sample characteristics: 23% had initially been assigned male, and those with atypical gender identity may have declined participation.

The current study was designed to provide systematic evidence relevant to current controversies regarding determinants of gender identity in two ways. The first goal was to extend the limited data on gender identity in females with CAH to examine effects of prenatal androgen exposure on gender identity. The second goal was to determine whether variations in gender identity are related to variations in degree of androgen exposure or genital appearance (degree of virilization and age at reconstructive genital surgery). To help put results in context, a small group of tomboy girls was also studied, because they behave in sex-atypical ways (25, 26) but have sex-typical genitals and no known disorders of sexual differentiation.

Subjects and Methods

Participants aged 3–18 yr included 43 girls with classical CAH due to 21-hydroxylase deficiency always reared as girls, 7 girls identified as tomboys, and 29 control girls (25 unaffected sisters and cousins of girls and boys with CAH, and 4 sisters of tomboys). Girls represented a range of socioeconomic backgrounds, and most were Caucasian. Girls with CAH and their relatives were participants in a study of social behavior and cognition that included girls and boys with CAH, recruited through university-affiliated pediatric endocrine clinics, and unaffected sibling and cousin controls. Other data from these subjects have been reported: 41% were studied previously when they were between 3 and 12 yr old (19, 27–30); adolescents (54% of the sample) have been described with respect to their concurrent activities (31). Girls identified as tomboys by their parents were recruited with their sisters through articles in local newspapers describing the study and recruiting participants. This is not a representative sample of tomboys, but they are included to place into context data from girls with CAH. Group differences in age reflect targeted recruitment of tomboys aged 4–11 yr. Mean ages in years (and *SD* values) are as follows: girls with CAH, 10.40 (4.26); female relatives of girls with CAH, 10.77 (4.65); tomboys, 8.15 (2.02); and sisters of tomboys, 7.18 (2.16). The research was approved by Institutional Review Boards at all cooperating institutions. Parents provided informed written consent for behavioral assessments and evaluation of medical records, and children provided written consent or oral assent for behavioral procedures.

Information from medical records was available for most girls with CAH, providing ratings for age at diagnosis and type of CAH (salt-

wasting or simple-virilizing; *n* = 26, 60% of the sample), genital virilization (*n* = 24, 56%), age at clitoral surgery (*n* = 19, 44%), and age at vaginoplasty (*n* = 20, 47%). Gender identity scores did not differ between girls with vs. without medical data. Most girls had a severe form of CAH; 77% were diagnosed before 6 months of age, and 88% had salt-wasting CAH. All had genital virilization at birth, but all were reared as girls from birth; Prader scores ranged from 1–4 (average, 3.0). Average age at clitoral surgery was 1.6 yr (range, 6 d to 10.9 yr; median, 1.1 yr). Average age at vaginoplasty was 4.6 yr (range, 8 months to 16.4 yr; median, 2.5 yr).

Given concerns about sampling bias in studies of intersex populations, it is important to describe recruitment and retention. On initial contact, over 90% of families agreed to participate in the study. Among girls who participated in the previous phase of this study, 40% did not participate in the current phase, primarily because of distance (we had moved), children's schedules, or family problems. Participating and nonparticipating girls with CAH did not differ on behaviors assessed previously or on disease characteristics that have been shown to be related to gender-role behavior and sexual orientation (22, 30, 32, 33).

Gender identity was measured with a nine-item interview (see *Appendix*), modified from one developed by Zucker (described in Ref. 17) to detect gender identity disorder. Our modifications, based on our experience with Zucker's interview in an earlier study of 60 tomboys (25), were intended to make the interview sensitive to subclinical manifestations of atypical gender identity and to address concerns of both girls and parents who found some of the questions (e.g. "Are you a boy?") overly provocative. Thus, our modified interview compares to Zucker's interview (17) in the following ways: three similar items assess preference for being a boy rather than a girl; items assessing cognitive confusion about gender identity were omitted because there was no variability in our prior study; two new questions assessing preference for feminine hairstyle and clothing specifically reflect diagnostic criteria for gender identity disorder of childhood (34); the other new items assess atypical gender identity not as extreme as persistent and intense distress about being a girl and include whether a girl would become a boy if it were "magically" possible, whether she would rather become a mother or a father, and whether she would choose a male identity if she could become someone else.

Each item was scored for sex-typicality (0, female-typical; 1, unclear or reflecting both female- and male-typical preferences; 2, male-typical). Scores on individual items were summed to yield a measure of gender identity ranging from 0 (complete female-typical identity) to 18 (complete male-typical identity). The items measure a single construct, as reflected by internal consistency reliability (coefficient α , 0.76), and item-total correlations (median, 0.45; details about the scoring procedures and psychometric properties of the gender identity interview may be obtained from S.A.B.). Girls were interviewed at home during a test session assessing several aspects of sex-typed behavior. Interviewers were aware of the status of some participants, but interviews were scored from interviewers' written records of subjects' responses by a clinical psychologist (J.M.B.) who had no knowledge of status, other identifying information, or other behavioral data.

The main comparison was of girls with CAH and control girls, but, as will be apparent, data from tomboys were also informative. For the total score, three groups were compared with one-way ANOVA and pairwise contrasts using (two-tailed) *t* tests; magnitudes of differences are reported in *SD* units, *d* (35). Group differences in variability were assessed with Levene's test. For individual items, groups were compared pairwise with χ^2 tests, with type I error set at 0.10 because of relatively small samples, especially of tomboys, and limited information on this topic.

Results

The groups differed in gender identity, as measured by total scores on the interview [Fig. 1; $F_{(2,76)} = 8.79$; $P < 0.001$]. (The pattern of results was similar when analyses were based on subsets of items, e.g. excluding items assessing preferred appearance.) On average, girls with CAH had scores intermediate between those of tomboys and control girls, but only 5 of 43 girls with CAH had scores outside the range of control girls. Girls with CAH had significantly higher scores than

control girls [$M = 5.81$; $SD = 3.41$, vs. $M = 3.69$; $SD = 2.55$, respectively; $d = 0.71$; $t(70) = 2.86$; $P < 0.01$], and significantly lower scores than tomboys [$M = 9.71$; $SD = 6.97$; $d = 0.71$; $t(48) = -2.37$; $P < 0.05$]. Group differences are not due to outliers. (One girl with CAH had received prenatal dexamethasone treatment and, thereafter, might be expected to be more sex-typical in her behavior than other girls with CAH. Her score was in the middle of the range for girls with CAH, and results were very similar when she was excluded.) Tomboys also had significantly higher scores than control girls [$d = 1.15$; $t(34) = 3.83$; $P < 0.001$], with 3 of 7 tomboys scoring outside the range of controls. Tomboys were also significantly more variable than girls with CAH [$F_{(48)} = 10.20$; $P < 0.01$] and control girls [$F_{(34)} = 19.87$; $P < 0.001$], who did not differ [$F_{(70)} = 0.97$; $P = \text{not significant}$]. Age was not significantly correlated with gender identity, either across subjects ($r = 0.13$; $P = \text{not significant}$) or within groups.

On individual items (Table 1), girls with CAH were generally similar to control girls, giving significantly more male-typical responses on only two of nine items. In contrast, tomboys were more likely than both control girls and girls

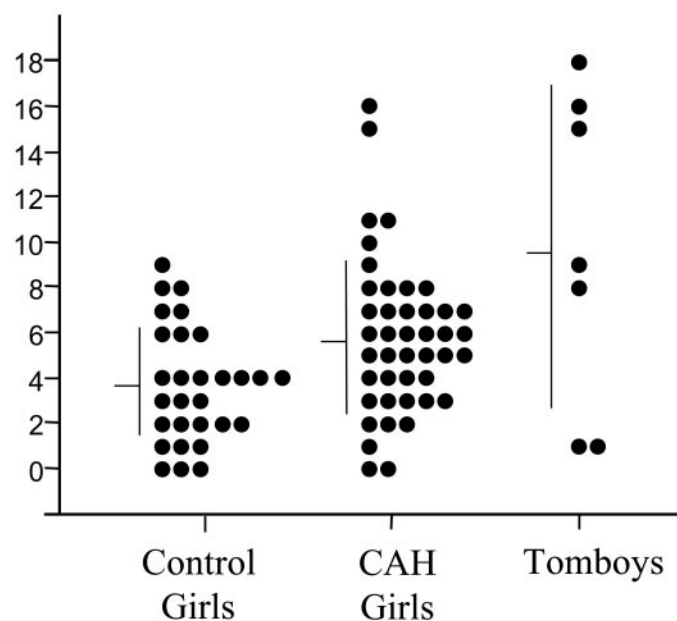


FIG. 1. Gender identity scores of individual control girls, girls with CAH, and tomboys. Horizontal lines represent means; vertical lines represent SD.

TABLE 1. Frequency of male-typical responses to items on the gender identity interview

	CAH girls (n = 43)	Tomboys (n = 7)	Control girls (n = 29)	Significant group differences
1. Prefers short hair	10 (23%)	3 (43%)	6 (21%)	
2. Does not like dresses	25 (58%)	3 (43%)	10 (35%)	cah/ctl ^a
3. Better to be boy	2 (5%)	2 (29%)	0 (0%)	cah/tb ^a , tb/ctl ^b
4. Not happy as girl	4 (9%)	3 (43%)	0 (0%)	cah/tb ^a , tb/ctl ^c
5. Wishes to be boy	12 (28%)	3 (43%)	5 (17%)	
6. Try boy for a while	23 (54%)	5 (71%)	11 (38%)	
7. Boy forever	1 (2%)	3 (43%)	0 (0%)	cah/tb ^c , tb/ctl ^c
8. Rather be father	2 (5%)	3 (43%)	0 (0%)	cah/tb ^c , tb/ctl ^c
9. Pretends male	9 (21%)	5 (71%)	2 (7%)	cah/tb ^b , cah/ctl ^b , tb/ctl ^c

cah, Girls with CAH; tb, tomboys; ctl, control girls.

Group differences on items scored 0, 1, or 2 tested by χ^2 , ^a $P < 0.10$; ^b $P < 0.05$; ^c $P < 0.01$.

with CAH to give male-typical responses, and, despite small sample size, differences were significant on five items.

Gender identity (total score) was not significantly associated with genital appearance or indicators of prenatal androgen excess in girls with CAH: $r(24) = 0.04$ with Prader rating, $r(19) = 0.05$ with age at clitoroplasty, $r(20) = -0.16$ with age at vaginoplasty, $r(26) = 0.11$ with salt-wasting status, and $r(26) = -0.20$ with age at diagnosis. For individual items, 3 of 27 correlations were significant, all between preference for short hair and indicators of prenatal androgen excess (Prader rating, CAH type, and age at diagnosis; r values about 0.40; $P < 0.05$). As with the total score, correlations of other individual items and genital virilization were low and nonsignificant, and some were not in the predicted direction; absolute r values ranged from 0.00–0.30 (median $r = 0.13$).

Of the five girls with CAH who had high scores on male gender identity (outside the range of typical girls), only one had an available Prader rating (it was 3). This created a challenge for testing the hypothesis that degree of genital virilization reflects brain masculinization as manifested in gender identity (13). Therefore, we assigned these girls a Prader score of 4 (Prader 5 is rare) and recalculated correlations between genital virilization and gender identity. As expected, correlations were higher than those calculated with actual data but were still nonsignificant. Correlations with estimated genital virilization were: gender identity total score, $r = 0.27$; "ever wishes to be a boy," $r = -0.06$; "if by magic, could be a boy forever," $r = 0.16$.

Discussion

These data are relevant to controversies about the management of children with intersex conditions, particularly regarding the effects on gender identity of prenatal androgens and genital appearance. Systematic evidence from this study confirms other reports in females with CAH to establish that development of male gender identity does not depend solely on prenatal androgen exposure or genital appearance. At the moderate levels to which females with CAH are exposed, early androgen exposure appears to have a modest effect on degree of gender identification, but not enough to produce gender dysphoria in the vast majority of cases. Girls with CAH had more male-typical scores than control girls on the gender identity scale, but only 5 of 43 girls with CAH had scores outside the range of scores of control girls. Importantly, the girls with atypical scores were not

those exposed to especially high prenatal androgen inferred from genitalia or other disease characteristics, indicating that genital appearance at birth does not reflect brain masculinization for gender identity and emphasizing the need to identify other causes of atypical gender identity. Results are consistent with other reports from females with CAH: most have female-typical gender identity, but a small number have cross-sex identification, and more than expected live comfortably as males. Those who live or identify as males are not necessarily those with the most severe genital virilization or highest prenatal androgen exposure (4, 20–22). Consistent failure to find associations between gender identity and degree of genital virilization or disease severity suggests that sex assignment in girls with CAH should not be based on genital appearance or inferred prenatal androgen levels (13).

Results also suggest that the development of female gender identity may not require normal-appearing genitalia. Gender identity was not significantly associated with age at genital reconstructive surgery, but caution is necessary in generalizing from this sample because most girls had surgery in early childhood and data were obtained from records and were not available for all girls.

Several methodological issues bear consideration in interpreting the results. First, medical data were not available for all girls, but those with data were similar to those without. Second, there was sufficient statistical power to detect a correlation of 0.50 between gender identity and genital appearance. It is likely that a larger correlation than this would be necessary for decisions about sex assignment to be based exclusively on genital appearance. Third, the gender identity interview itself was not validated in a sample of females with gender identity disorder, but scores showed variation, particularly in tomboys, and many items are similar or identical to those used by others to detect gender dysphoria (17, 34). Results of item analyses and group comparisons on subsets of scores suggest that the interview does measure a single construct. Fourth, to increase statistical power, type I error was set higher than is usual for analyses with individual items and was increased by the lack of correction for multiple statistical comparisons, but differences between girls with CAH and control girls were still generally not significant. The increased statistical power to detect group differences increases confidence that gender identity is not different between girls with CAH and typical girls in the population. Fifth, it is difficult to generalize about tomboys given sample size and recruitment, but their data support the validity of the gender identity measure, and underscore the complex origins of gender identity. Tomboys had the most atypical responses, but were reared as girls and had no obvious signs of sex-atypical hormone exposure.

The study may also shed light on determinants of gender identity in individuals with other causes of intersexuality in whom there is considerable variability and plasticity in gender identity (2, 11). Sex assignment is based on genital appearance and, in cases of intersexuality, on sex chromosomes, inferred androgen exposure, and reproductive potential. Most individuals experience concordance between internal feelings and self-concept on the one hand, and assigned sex on the other. For most people, concordance is sufficient to accept assigned sex; those who do not accept

assigned sex are gender dysphoric (36). We suggest that graded variations in identification with the assigned sex reflect the recognition by some people (such as girls with CAH and tomboys) that aspects of their behavior, preferences, or feelings are not typical of members of their sex. This variation would be sufficient to cause conflict about gender identity only in unusual and extreme cases. Identifying the factors that differentiate the majority of females with CAH who have female gender identity from the small minority who have male gender identity might aid in predicting gender identity in the difficult cases of intersexuality (e.g. partial androgen insensitivity) and illuminate causes of extremely atypical gender identity. These factors may include hormones (e.g. higher sensitivity to androgen than typical for girls with CAH, low levels of ovarian hormones), genes (especially Sry), social environment (e.g. inconsistent rearing environment), or individual characteristics (e.g. felt pressure to have gender identity conform to gender role).

These results reinforce the complexity of androgen effects on human behavior. It is clear that females with CAH are sex-atypical in some ways and not others. Even when they differ from control females, the magnitude of the difference varies across behaviors (37). Variability in behavioral effects of androgens is also found in other species, and likely reflects characteristics of androgen exposure (e.g. dose, timing, form of androgen, interactions with other hormones) and modification by the social environment (38–41). This means that it is not possible to predict across behaviors, particularly from gender-role behavior such as toy play to gender identity or sexual orientation, or to predict behavior from any single factor, such as genital appearance, androgen exposure, or rearing environment. The complex origins of gender identity and gender-related behaviors need to be considered in the medical management of children with intersex conditions.

Appendix: Gender Identity Interview

"I want to ask you some questions about how boys and girls are different. Remember there are no right or wrong answers to these questions. We just want to know how you feel about some of these differences."

1. "Some girls like to have long hair, other girls don't, and some girls don't care. Which do you like better?" (Long hair, Short hair, Doesn't care)

2. "Some girls like to wear dresses, some girls don't like to wear them, and some girls don't care. Do you like to wear dresses?" (Yes, No, Doesn't care)

3. "Some people think it's better to be a girl. Some people think it's better to be a boy, and some people think there are some good things about being girls and other good things about being boys. What do you think?" (Better to be a girl, Better to be a boy, Good things about each)

If child answers "girl" or "boy," ask: "What do you think is better about being a _____?"

Then ask: "Is there anything good about being a (the opposite of the first answer)?"

If child answers "both," ask: "Tell me some good things about being a girl." Then ask: "Tell me some good things about being a boy."

4. "Some girls are always happy being girls, but sometimes some girls wish that they could be boys. Are you happy being a girl?" (Yes, No)

If the child answers she is not always happy, ask: "Tell me about a time when you are not happy being a girl."

5. "Do you ever wish you could be a boy?" (Yes, No)

If the child answers "yes" ask: "Tell me about a time when you wish you are a boy."

6. "If someone knew a magical way to turn you into a boy, but just for a little while, would you want to try it?" (Yes, No)

7. Asked if the child answers “yes” for #6; otherwise the answer is assumed to be “no.”

“If someone knew a magical way to turn you into a boy forever, so that you would never be a girl again, would you do it?” (Yes, No)

8. “When you grow up would you rather be a mother or a father?”

If the child hesitates or doesn’t know, ask: “Do you want to be like your mother or like your father?” (Mother, Father, Both, Neither, Don’t know)

9. “Some people like to imagine or pretend that they’re someone else. If you could pick one person to become, besides yourself, who would it be?”

If necessary, child is questioned to determine the sex of the person.

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