EXHIBIT N

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Gender Dysphoria and Gender Change in Chromosomal Females with Congenital Adrenal Hyperplasia

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This article reviews the literature on studies and case reports on gender identity and gender identity problems, gender dysphoria, and gender change in chromosomal females with congenital adrenal hyperplasia, raised male or female. The large majority (94.8%) of the patients raised female ($N = 250$) later developed a gender identity as girls and women and did not feel gender dysphoric. But 13 (5.2%) patients had serious problems with their gender identity. This percentage is higher than the prevalence of female-to-male transsexuals in the general population of chromosomal females. Among patients raised male, serious gender identity problems were reported in 4 (12.1%) out of 33 patients. From these observations, we conclude that the assignment to the female gender as a general policy for 46,XX patients with CAH appears justified, even in severely masculinized 46,XX newborns with CAH (Prader stage IV or V).

KEY WORDS: congenital adrenal hyperplasia; gender identity; gender dysphoria.

INTRODUCTION

Congenital adrenal hyperplasia (CAH) is a disorder in which the adrenal glands produce excessive amounts of androgens. In chromosomal females, the classical (prenatal-onset) form of the disorder causes genital masculinization, ranging from mild clitoral enlargement to complete fusion of the labioscrotal folds with a phallic urethra (Speiser & White, 2003). In the large majority of girls, the disorder is diagnosed through (1) a work-up following the identification of genital ambiguity at birth, (2) neonatal screening in the first weeks after birth (Therrell, 2001; Van der Kamp et al., 2001), or (3) a medical work-up when the child develops an electrolyte crisis secondary to salt loss in the first two weeks after birth, or undergoes progressive virilization during infancy and early childhood (Speiser, 2001). Girls with nonclassical CAH are born with normal female genitalia, and the disorder becomes symptomatic only during childhood or later (White & Speiser, 2000).

In Western industrialized countries, 46,XX neonates with CAH, if correctly diagnosed, are almost always assigned to the female gender because they have ovaries, a uterus, and a vagina, and will feminize normally in puberty and have the potentially for fertility if they are on adequate glucocorticoid replacement therapy. Sometimes female neonates are born with severely masculinized genitals and are assigned the male gender, particularly if the disorder is not recognized. When, in such cases, the correct diagnosis is made after the neonatal period, the prevailing policy advises gender reassignment, based on the usual medical course described above, the fertility potential, and the assumption that in early childhood gender identity is not yet stable. In relatively rare cases—mostly in patients with the simple virilizing type and the most severe degree of masculinization (Prader stage V)—the diagnosis is not made within the first 18 months of life but at a much later age. For these patients, there is no consensus about gender reassignment. Some undergo a gender reassignment, but others do not. The decision not to change gender may be made according to the recommendation made by

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Money, Hampson, and Hampson (1955). Based on a review of all data available in the literature and the evaluation of patients seen at Harvard University and Johns Hopkins Hospital, Money et al. (1955) concluded that gender assignment after the first 2 years of life or so led to increasing adjustment problems (see also Money, Hampson, and Hampson, 1957). Money's guidelines for patient management explicitly included that later gender reassignment should be based on careful evaluation of the child's behavior and gender wishes (Money, 1968). In most children who are assigned the female gender, the genitals are surgically feminized in order to support the development of a stable female gender identity, and to provide the capacity for heterosexual intercourse later in life.

Classic 21-hydroxylase deficiency is detected in approximately 1 in 16,000 births in most populations (Speiser & White, 2003). As classical CAH is the most frequent intersex disorder in 46,XX individuals, it has been possible to carry out studies on comparatively large groups of patients. Studies on gender behavior in patients raised female have shown that, as a group, they show variably masculinized behavior in regard to play, playmates, toys and activities in childhood, leisure time and other activities in adolescence and adulthood (Berenbaum, 2003; Collaer & Hines, 1995; Zucker, 2005). Dittmann (1992) observed slightly but significant more masculine positions and body movements in CAH females.

The prevailing policy of gender assignment of all chromosomal females with classical CAH to the female gender is currently under debate. On theoretical grounds, Diamond and Sigmundson (1997) proposed that chromosomal females born with severely masculinized genitals (Prader stage V) should be assigned the male gender. They assumed that the high levels of intrauterine androgens lead to a higher degree of brain masculinization and hence to an increased risk of gender dysphoria in females. In order to find arguments whether the prevailing gender assignment policy should be continued or replaced by Diamond and Sigmundson's proposal, we reviewed the literature on studies and case reports on gender identity and gender identity problems, gender dysphoria, and gender change in chromosomal females with CAH, assigned either to the female or to the male gender.

METHOD

This review is based upon the most frequently cited publications in the English-language literature from 1950 onwards in which gender development has been studied. We searched Medline and PsycLit with the search words congenital adrenal hyperplasia, CAH, gender, and author names. In addition, we consulted the reviews by Chan-Cua, Freidenberg and Jones (1989), Collaer and Hines (1995), and Zucker (1999).

We did not consult the literature before 1950 because hydrocortisone treatment was not available until that year and the majority of patients died in infancy. Furthermore, we selected studies in which there were no doubts about the diagnosis of CAH, and in which the children studied were at least two years old because it is not known whether gender identity or gender dysphoria exists in younger children.

We used different criteria in the selection of studies of 46,XX children raised female and 46,XX children raised male. For children raised female, we only included studies in which psychological measurements and/or psychiatric diagnostic instruments had been used in the assessment of gender outcomes. Most of these studies included female non-CAH controls. By contrast, the available studies or case reports on 46,XX children raised male commonly did not employ psychological measurements and/or psychiatric diagnostic instruments. In some studies, a psychiatrist had been consulted; in others, the judgement on gender development seemed to have been made by the pediatrician or the parents. These latter judgements might have been influenced by the fact that both the pediatrician and the parents are or had been directly involved in the original gender assignment decision. We, therefore, decided to include all studies in children raised male which included some statements about the gender development of these children, even if it was not formally assessed. None of the studies of male-raised cases included control subjects.

RESULTS

Table I summarizes the findings from studies of 46,XX individuals assigned and raised female ($N = 250$). The majority of the females suffered from classical CAH and had mild to severe masculinization of their external genitals. Thirty-two had been assigned male at birth, of which 28 were reassigned as females in the first 19 months of life. Slijper's (1984) study included one girl with simple virilizing CAH who was reassigned from male to female at age 3.5 years. In this girl, the disorder was recognized late. The decision to reassign gender was made because the child preferred to be a girl and psychological assessment revealed that she was developing a female gender identity. Wölfle et al.'s (2002) sample included three patients who had undergone a physician-imposed gender reassignment to female at ages 7–9 years. One of them accepted this change and did not have many social problems. Another girl accepted the female gender but had to deal with a poor acceptance by her social environment. The third girl did

Table I. Gender Identity in 46,XX Individuals with Congenital Adrenal Hyperplasia Raised Female

General information					Gender history			Latest report			
Reference	Country	Syndrome specifications	Severity of masculinization at birth	N[a]	GA at birth	Phys GRA at age	Pat GRA at age	Age[b]	Lives as M/F	Identifies as M/F/other	Gender dysphoria
Ehrhardt et al. (1968)[c]	USA	n.i.	Prader IV 7p	15	M: 7p F: 8p	before 8th month	None	5–16	F	F[d] 1p confused about her femininity	No[f]
Ehrhardt and Baker (1974)[c]	USA	SW 9p SV 8p	n.i.	17	n.i.	n.i.	None	4–20	F	F[d]	No
McGuire, Ryan, and Omenn (1975)[c]	USA	n.i.	n.i.	15	n.i.	n.i.	n.i.	7–20	F	F	n.i.
Slijper (1983, 1984)[c]	NL	SW 13p	Prader II 4p Prader III 3p Prader IV 6p	24	M: 5p F: 8p	before 13th month	None None	7–17	F	SW: F[d,e]	No[f]
		SV 11p	Prader II 8p Prader III 1p Prader IV 2p		M: 1p F: 10p	at age 3.5[g]				SV: F	
Hurtig and Rosenthal (1987)[c]	USA	n.i.	n.i.[h]	9	n.i.	n.i.	None	13–21	F	F[e]	No
Dittmann et al. (1990)[c]	GER	SW 13p SV 20p	Prader II–V	34	n.i.	n.i.	None	11–41	F	F	F
Meyer-Bahlburg et al. (1996)	USA	SW (p2)[i]	Markedly	1	M	1st month	Adulthood	35	M	M	Yes
		SW (p4)[i]	Markedly	1	F		Adulthood	30	M	M	Yes
		SV (p1)[i]	Markedly	1	M	1st month	Adulthood	28	M	M	Yes
		11βOH (p3)[i]	Markedly	1	F		Adulthood	38	M	M	Yes
Zucker et al. (1996)[c]	CAN	SW 19p SV 12p	n.i.	31	n.i[j]	1–2 month	None[k] after birth	18–40	F	F[d]	No
Slijper et al. (1998)	NL	n.i.	Severely 8p Ambiguous 2p	18	M: 8p F: 10p	6p at 0–3 month 2p at 6 month	None	2–27	F	F: 16p M: 2p[l]	No 16p Yes 2p[l]
Wölfle et al. (2002)	GER + SW	SW 12p SV 2p 11βOH 2p	Prader V 9p	9	M	6p patients in 1–19 month 3p patients at age 7–9year	None None	8–49	F F	F F: 2p (p8[m],p9)[i] M: 1p (p7)	No No Yes

Table I. Continued

General information					Gender history			Latest report			
Reference	Country	Syndrome specifications	Severity of masculinization	N[a]	GA at birth	Phys GRA at age	Pat GRA at age	Age[b]	Lives as M/F	Identifies M/F/other	Gender dysphoria
Berenbaum and Bailey (2003)[c]	USA	SW 35	Prader I–IV	43	F	No	None	3–18	F	F	No
		SV 8	average Prader III								
Hines et al. (2004)[c]	UK	SW 14	n.i. n.i. 2	16	n.i.	n.i.	None	18–44	F	F[d]	No 11p Yes 5p[n]
Meyer-Bahlburg et al. (2004)[c]	USA	SW 13 SV 2	Prader I 1p Prader II 1p Prader III 2p Prader IV 4p Prader IV/V 1p n.i. 6p	15	n.i.	n.i.	None	5–12	F	F	No

Notes. GA: gender assignment at birth; phys GRA: physician-imposed gender reassignment; pat GRA: patient-initiated gender reassignment; m/f/other: male, female or other; n.i.: no information; SW: CAH salt wasting type, SV: CAH simple virilizing type; p: patient(s); USA: United States; NL: The Netherlands; CAN: Canada; GER + SW: Germany and Switzerland.

[a] *N* only refers to the number of CAH patients who participated in the study (excluded are the number of control subjects and the total number of patients who had been asked to take part in the follow-up study).

[b] The age at the latest report is presented because in many studies we were unable to find out at what ages the child had been seen before but descriptions made clear that many patients had been admitted to several hospitals before.

[c] Comparison of CAH patients and matched controls or family controls (sisters, cousins, mothers).

[d] These females identified themselves as females, but on measurements they indicated they felt less typically female compared to controls.

[e] Showed a less-female-typical identification on the Draw-a-Person test.

[f] These girls did not feel gender dysphoric but felt less content with being female or more often expressed the wish to have been born male.

[g] This decision was based on a careful assessment of the patient's behavior and preferences.

[h] Four patients had virilization at the time of the study; in three of them, this was due to non-compliance with medication.

[i] In the publication referred to as patient no. 1,2, etc.

[j] Information is given for the total sample but not for the patients who entered the follow-up study.

[k] The total study group (which is larger than the 31 subjects who participated in this study) comprised one female who changed gender to male at age 19. He refused to take part in the study.

[l] Onset of GID at age 3 and 4.

[m] This patient accepted the female gender, but acceptance by her social environment was poor.

[n] 5 females indicated they had wished to be a person of the other sex in the past 12 months, 4 endorsing "some of the time" and 1 endorsing "about half of the time."

not accept the reassignment to female and also had to deal with people in her social network who did not accept the change.

The females in the studies by Ehrhardt, Epstein, and Money (1968), Ehrhardt and Baker (1974), Slijper (1984), Zucker et al. (1996), Slijper, Drop, Molenaar, and de Muinck Keizer-Schrama (1998) and Hines, Brook, and Conway (2004) identified themselves as females, but on measurements they indicated that they felt less typically female compared to the control group.

Of the 250 CAH females who participated in these investigations, 237 did not show symptoms of gender dysphoria. However, 13 females reported gender problems: one female was confused about her female identity, 5 indicated that they sometimes had wished to be a person of the other sex in the past 12 months, and 7 experienced gender dysphoria. Four (30%) of these 13 females experienced such a severe dysphoria that they wanted to change their gender (Meyer-Bahlburg et al., 1996).

Table II shows the findings from studies of 46,XX individuals raised male ($N = 33$). We found 12 studies on gender identity in 46,XX individuals raised male. Most studies included only one or two individuals, except those of Rösler and Leiberman (1984), Sripathi, Ahmed, Sakati, and al-Ashwal (1997), and Wölfle et al. (2002). At the time of study, 4 (12.1%) out of these 33 chromosomal females raised male identified themselves as female (1 patient) or were gender dysphoric (3 patients).

These data suggest that gender problems seem to occur more often among CAH patients raised male than among those patients raised female. However, the difference between male and female raised patients was not statistically significant ($p = .12$, Fisher's Exact Test, two-tailed).

DISCUSSION

The studies reviewed in this article show that, despite the masculinized gender behavior, the majority (94.8%) of chromosomal females with CAH raised female identify themselves as females and live in their assigned gender (Table I). Only a few of them, 13 (5.2%) out of 250, experienced gender dysphoria. One of these patients had originally been raised male but underwent physician-imposed gender reassignment to female at age 7. It has been reported before that such a late gender change is associated with an increased risk of gender dysphoria (Wölfle et al., 2002). Four (30%) of the 13 gender dysphoric patients experienced such a severe dysphoria that they initiated a gender change in adulthood. In the total group of evaluated patients, the percentage of females who changed gender was 1.6% (4 out of 250). In addition, a fifth patient who changed gender was known from the data base of Zucker et al. (1996). The number of females who desired a gender change is larger than expected, given the baseline prevalence of female to male transsexualism in the general population of chromosomal females (1:30,400; Bakker, van Kesteren, Gooren, & Bezemer, 1993). The females who changed their gender did not differ from other women with CAH with respect to severity of genital masculinization or levels of prenatal androgen exposure (Meyer-Bahlburg et al., 1996; Zucker et al., 1996). Based on these findings, it has been concluded that an apparently stable gender identity in childhood does not necessarily preclude gender dysphoria and gender change later in life nor does a successful gender reassignment in childhood rule out later gender problems (Meyer-Bahlburg et al., 1996; Zucker et al., 1996).

Four chromosomal females raised male experienced gender dysphoria and 29 did not. With respect to the occurrence of gender dysphoria, there was no significant difference between 46,XX CAH patients raised male and patients raised female. The information provided in the literature is often not specific enough to permit a conclusion regarding the question whether the 46,XX patients raised male (Table II) had more masculinized genitals at birth compared to those raised female (Table I). It appears that all of these children had been assigned the male gender initially before their condition was correctly diagnosed. In the case of some 46,XX children raised male, their 46,XX karyotype became known within their first two years of life, but at that point, the parents did not want to reassign their child to the female gender. Our findings indicate that, compared to the general population, gender dysphoria is more often seen among these patients, either raised male or female. The large majority of patients, however, do not experience gender dysphoria. These observations confirm the conclusion by Hines (2004) that gender identity development, at least in patients with classical CAH, is remarkably flexible.

In regard to Diamond and Sigmundson's proposal (1997), the sample of Wölfle et al. (2002) is of interest because it only includes children born with completely masculinized external genitalia (Prader stage V). This study included 16 patients all assigned male at birth. Seven of them had been raised male (Table II), 6 had been raised female from 1 to 19 months after birth onwards, and 3 had been raised male in the first 7–9 years of life. Among the 7 male raised patients, one developed a disturbed gender identity (Table II). Of the 6 patients who had undergone early gender reassignment, none developed gender identity problems

Table II. Gender Identity in 46,XX Individuals with Congenital Adrenal Hyperplasia Raised Male

General information					Gender history			Latest report			
Reference	Country	Syndrome specifications	Severity of masculinization	N[a]	GA	Phys GRA at age	Pat GRA at age	Age[b] at study	Lives as M/F	Identifies M/F/Other	Gender dysphoria
Wilkins (1957) and Jones and Scott (1958)[c]	USA	SW	n.i.	1	M	—	—	7½	M	Psychiatric advice to continue the male role	
Peris (1960)[c]	USA	n.i.	n.i.	1	M	—	—	18	M	M	No
Madsen (1963)[c]	USA	n.i.	n.i.	2[d]	M	—	—[e]	35	M	F	Yes
					M	—	—	30	M	M	n.i.
Gillenwater, Wyker, Birdsong, and Thornton (1970)[c]	USA	n.i.	n.i.	1	M	—	—	7	M	M	n.i.
Redman and Gould (1972)[c]	USA	SW	n.i.	1	M	—	—	6	M	Psychologically a boy	
Money and Daléry (1976)[c]	USA	SW	Prader IV or V	1	M	—	—	adult	M	M/Masculine personality	
		n.i.		2[d]	M	—	—	adult	M		
Rösler and Leiberman (1984)	IS	11βOH	severe	8	M	—	—	17–33	M	6 M 1 ambivalent 1 not adjusted as a male	No
Wyatt, Chasalow, Granoff, and Blethen (1987)	USA	SV	Prader V	1	M	n.i.	—	3½	n.i.	M	No
Chan-Cua et al. (1989)[c]	USA	SW (p1)[f]	Prader V	1	M	—	—	8¼	M	M	No
		SV (p2)[f]	Prader V	1	M	—	—	4½	M	M	No
Sripathi et al. (1997)[c]	USA	n.i.	n.i.	6	M	—	—	3–16	M	Well adjusted to boyhood	
Wölfle et al. (2002)[c]	GER + SW	n.i.	Prader V	7	M	—	—	8–49	M	M: 6p 1p (p2)[g,h]	No 1p (p2)[g]

Notes. GA: gender assignment at birth; phys GRA: physician-imposed gender reassignment; pat GRA: patient-initiated gender reassignment; m/f/other: male, female or other; Pso measurement/psa diagnosis: psychological measurements or psychiatric diagnostic evaluation has been applied; n.i.: no information; USA: United States; IS: Israel; GER + SW: Germany and Switzerland; SA: Saudi Arabia.

[a] *N* only refers to the number of evaluated CAH patients.

[b] The age at the latest report is presented because in many studies or reports we were unable to find out at what ages the child had been seen before but descriptions made clear that many patients had been admitted to several hospitals before.

[c] In these children, the disorder was either recognized late or the parents did not want to reassign the child's gender. In two brothers (Sripathi et al., 1997), the disorder was recognized at birth but the parents insisted on male upbringing.

[d] Brothers.

[e] This patient sought help because he wanted to become a woman. From age 26 on, when he started to menstruate, he was convinced that he was a woman.

[f] The authors describe 3 patients raised as males. In one patient, his identification as a male can be understood from the gender confusion he suffered when he developed breasts at age 11–12 and his refusal to follow his doctor's advice to live as a girl. The patients are further described having masculine personalities.

[g] In the publication, referred to as patient No 2.

[h] According to the author's description, he has a "disturbed gender identity."

(see Table I). Of the 3 children who had undergone late gender reassignment, only one accepted her female gender and did not experience social stigmatization. For the other two children, the reassignment decision appeared to be unfortunate: one child kept identifying herself as a male whereas the other child accepted her new gender identity but suffered from social stigmatization as her new gender identity was not accepted by others. From their findings, Wölfle et al. (2002) concluded that physician-imposed gender reassignment late in childhood should be avoided. This conclusion is in agreement with the original recommendations by Money et al. (1955). Wölfle et al. (2002) plea for neonatal screening and recommend gender reassignment (if appropriate) in initially undiagnosed children under 19 months of age as soon as the correct diagnosis has been made. The study is an important one, and should be repeated in other 46,XX individuals raised in the male gender because such studies broaden our knowledge of gender development and gender problems in these children (Meyer-Bahlburg et al., 2004).

Berenbaum and Bailey (2003) investigated the effects on gender identity of prenatal androgens and genital appearance but did not observe a relationship between degree of genital masculinization and prevalence of gender identity problems or gender dysphoria. They pointed out that the relationship between genital masculinization and brain masculinization may not be proportional and that caution is needed to conclude that gender identity is determined primarily by prenatal androgen exposure. A similar conclusion was drawn by Meyer-Bahlburg et al. (2004), who observed masculinized gender behavior but no gender dysphoria in 5–12 year-old CAH girls. A relationship between prenatal androgenization and masculinization of gender behavior has been observed in many studies, but not in all (Collaer & Hines, 1995). Published correlations of gender related behavior with Prader stage of genital masculinization have been inconsistent, ranging from .00 to .68 (Meyer-Bahlburg, 2001). The studies reviewed in this article indicate no relationship of prenatal androgenization with masculinization of core gender identity.

The difference between gender behavior and gender identity in their association with prenatal androgenization suggests that the underlying mechanisms establishing gender behavior and gender identity are not quite the same. With respect to gender identity, in addition to prenatal androgenization, genetic factors may play a role (Arnold, 2002; De Vries & Simerly, 2002). Another explanation could be that, in these patients, social factors influence the development of gender identity more strongly than the development of gender-related behavior (Meyer-Bahlburg et al., 2004).

From the findings in the studies described in this article, one could optimistically conclude that the present medical and psychosocial management of these patients is not such a bad one. However, the data show that, although many patients adjust well to their lives, the proportion of patients who experience significant gender problems is higher than expected. In order to gain a better understanding of these gender problems, future studies should include specific assessments of gender identity and gender dysphoria that will permit better data on prevalence, characteristics and severity of gender problems. As many gender problems seem to arise in late adolescence and adulthood, more of these studies should be performed in patients 17 years of age and older.

We also need to address the problem of patient representativeness. Patient advocacy groups (ISNA, 2003) are dissatisfied with the present policy. They point out that, particularly in adulthood, many patients encounter gender-related and psychosexual problems, which some attribute to the medical intervention. Zucker et al. (1996) mentioned the well-known problem in clinical research that patients dissatisfied with the services they received may be resistant to participation in follow-up studies. Consequently, the participating patients are somewhat more likely to include satisfied ones. This may lead to an underrepresentation of problematic outcomes and, perhaps, an inadequate revision of treatment policies. It is, therefore, very important that also patients who are dissatisfied realize the importance of participating in such follow-up studies, and that investigators make special efforts to facilitate their participation.

We agree with Berenbaum (2003) that it is not necessary to make substantial changes in our medical and psychosocial management. However, the encountered gender-related problems deserve more attention. Our knowledge is limited and we are unable yet to predict which patients will develop gender identity problems and which will not. Patients and their parents should be informed about this, and psychological help should be offered to help these patients and to understand the specific nature of their problems. Only the (older) patient him/herself can judge whether the gender s/he has been assigned to is compatible with his/her overall experience of the self in the given social environment.

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