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Fetal Feminization and Female Gender Identity in the Testicular Feminizing Syndrome of Androgen Insensitivity[1]

Daniel N. Masica, A.B.,[2,3] **John Money,** PH.D.,[2,4] **and Anke A. Ehrhardt,** PH.D.[2]

Interview data on aspects of sex and eroticism from a sample of 10 chromosomally male (XY) patients with the complete testicular feminization (androgen insensitivity) syndrome and from a sample of 23 patients with the late-treated adrenogenital syndrome showed marked differences. Homosexual experiences and/or dreams were lacking in the androgen-insensitive group as compared with the adrenogenital group ($p \leqslant 0.01$). The androgen-insensitive group reported lower frequency of sexual arousal from visual stimuli than did the adrenogenital group ($p \leqslant 0.05$). Findings on different aspects of sexual behavior suggest a tendency for the androgen-insensitive patients as a group to have a lower sex drive, to be less keenly aware of their sex drive, to be less assertive in heterosexual relations, and to be less versatile in coitus than the adrenogenital patients. No case of exclusive lesbianism, transsexualism, or transvestism was reported from either patient group. Although the two groups differed in sexual and erotic behavior, both were within the range of what in our culture is accepted as feminine. Nonetheless, the androgen-insensitive patients conformed more closely to the conventional feminine stereotype. Regarding explicit satisfaction with female sex role and with cosmetic and clothing interests, the androgen-insensitive group was characteristically feminine. Findings on the Draw-a-Person Test and the Guilford–Zimmerman Temperament Survey are compatible with the results of normal females, but are in sharp contrast with those of normal males. Interview and psychometric data thus concur in showing the androgen-insensitive patients to be unmistakably feminine in behavior and outlook. Their femininity is best conceived of as a product of hormonal nonandrogenization, prenatally and later, in combination with the social experiences of rearing and development, after initial assignment as a girl.

INTRODUCTION

This is the third in a series of articles dealing with the discrepancy between female gender identity and hormonal sex *versus* male genotypic and gonadal sex in the

[1] Supported in research by Grants 5K03-HD18635 and 2R01-HD00325 USPHS.

[2] Department of Psychiatry and Behavioral Sciences and Department of Pediatrics, The Johns Hopkins University School of Medicine, Baltimore, Maryland.

[3] D. N. Masica was a Henry Strong Denison Scholar in Medical Research, The Johns Hopkins University, 1969–70.

[4] Requests for reprints should be directed to John Money, Ph.D., Phipps 400, The Johns Hopkins Hospital, Baltimore, Maryland 21205.

testicular feminizing syndrome of androgen insensitivity (Money *et al.*, 1968; Masica *et al.*, 1969).[5] Surveys and compiled case reports on the syndrome of testicular feminization have included for the most part only anecdotal impressions on psychosexual orientation, level of libido, and conformity to conventions of femininity (Morris, 1953; Hauser *et al.*, 1957; Morris and Mahesh, 1963).

PURPOSE

The purpose of this paper is to present a systematic examination of the data on gender identity and on aspects of sex and eroticism, obtained in interviews and from psychometric testing, from 10 patients diagnosed as unequivocal cases of testicular feminization

SAMPLE SELECTION AND DESCRIPTION

The 10 patients in the sample (Table I), all with the complete syndrome of testicular feminization, constitute virtually a census of patients, in late adolescence and adulthood, followed in the pediatric endocrine clinic and the gynecology service between 1951 and 1968.[6] The lower age at time of last psychologic follow-up was set at 16 years, on which basis eight juveniles did not qualify. Only two adult women were known to have been missed completely: one because of distance and the other because of privacy problems.

[5] Clinical symptoms and signs are discussed in Money *et al.* (1968).

[6] These 10 composed the total sample of the first paper and two-thirds of the sample of the second paper in this series on the psychological aspects of the androgen insensitivity syndrome (Money *et al.*, 1968; Masica *et al.*, 1969).

Table I. Descriptive Data on 10 Patients with the Androgen Insensitivity (Testicular Feminizing) Syndrome

Patient	Date of birth	Race	Marital status	Profession	Age at first estrogen treatment (years)	Taking of estrogen treatment
A[a]	12/19/36	White	Married	Housewife	21 2/12	Regularly
B[a]	11/25/39	White	Married	Housewife	20 4/12	Regularly
C[a]	9/28/49	White	Married	Housewife	15 10/12	Regularly
D[b]	1/24/48	Negro	Single	Secretary	17 8/12	Occasionally forgets
E	7/20/47	White	Married	Housewife (part-time cashier)	19 6/12	Regularly
F	11/8/53	White	Single	Student (11th grade)	13 6/12	Deliberately refuses to take treatment
G[b]	8/9/40	Negro	Single	Assistant librarian	18 7/12	Regularly[c]
H	8/15/44	White	Single	Professional model	13 0/12	Regularly[c]
I	7/12/52	White	Single	Student (12th grade)	11 11/12	Regularly
J[b]	1/4/47	Negro	Single[d]	Secretary	17 9/12	Regularly[c]

[a] Three sisters.

[b] Three cousins (the only Negro patients in the sample).

[c] Initial misconception as to purpose of estrogen pills; recommenced regular intake of treatment after explanation in our psychohormonal unit.

[d] At the time of last follow-up, patient was engaged.

Three of the 10 patients were sisters, and three others, each of separate parentage, were first cousins. Seven patients were white. The three cousins were black. Four of the 10 patients were married, and one was engaged. The two oldest married patients had two adopted children each.

Age at initial estrogen treatment varied from 11 $\frac{11}{12}$ through 21 $\frac{2}{12}$ years, the mean being 16 $\frac{11}{12}$ and the median 17 $\frac{8}{12}$ years. Five of the patients had taken their estrogen treatment regularly from the first treatment onward (Table I). Three other patients initially had misconceptions as to the purpose and usefulness of their estrogen pills and were taking them sporadically. These three changed to regular estrogen intake after explanation by the authors. One other patient was lax, occasionally forgetting to take her pills. The remaining patient rejected estrogen treatment (see below).

Additional data, including age at time of diagnosis and age at gonadectomy, are found in the already published two papers in this series.

The 23 patients with the late-treated adrenogenital syndrome, used for contrast

Table II. Interview Data on Aspects of Sex and Eroticism in 10 Patients with the Testicular Feminizing Syndrome Contrasted with 23 Patients with the Late-Treated Adrenogenital Syndrome

Type of experience	Frequency and percent (in parentheses) of total	
	Testicular feminization (androgen insensitive) group (N = 10)	Late-treated adrenogenital group (N = 23)
A. Sexual practices		
1. Heterosexual relations		
a. Frequent heterosexual and no homosexual experience	8 (80)	11 (48)
b. Frequent heterosexual and occasional homosexual experience	0	2[a] (9)
c. Frequent heterosexual and frequent homosexual experience	0	2 (9)
d. Very limited heterosexual and no homosexual experience	0	1 (4)
2. Homosexual relations exclusively		
a. Frequent	0	0
b. Very limited	0	0
3. No experience in sexual relations		
a. Masturbation only	0	4 (17)
b. No masturbation	2 (20)	2 (9)
c. No information	0	1 (4)
B. Erotic dreams and fantasies *vis-à-vis* actual sexual experience		
1. Heterosexual experience exclusively		
a. Homosexual dreams	0	5 (22)
b. No homosexual dreams	8 (80)	7 (30)
2. Masturbation experience only		
a. Only heterosexual dreams	0	2 (9)
b. Hetero/homosexual dreams	0	2 (9)
c. Only homosexual dreams	0	0
3. No sexual experience		
a. Only heterosexual dreams	1 (10)	0
b. Hetero/homosexual dreams	0	0
c. Only homosexual dreams	0	0
d. No dreams	1 (10)	2 (9)
e. No information	0	1 (4)

Table II—contd.

Type of experience	Frequency and percent (in parentheses) of total	
	Testicular feminization (androgen insensitive) group ($N = 10$)	Late-treated adrenogenital group ($N = 23$)
4. Bisexual experience		
a. Hetero/homosexual dreams	0	3 (13)
b. No information	0	1 (4)
C. Libido		
1. Patient's own estimate of level		
a. Above average	2 (20)	11 (48)
b. Average	6 (60)	8 (35)
c. Below average	0	1 (4)
d. No information	2 (20)	3 (13)
2. Climax experience (all types of sexual practices)		
a. Always	2 (20)	2 (9)
b. Most of the time	6 (60)	14 (60)
c. Never	1 (10)	2 (9)
d. No information	1 (10)	5 (22)
3. Role in heterosexual relations		
a. Predominantly initiating	0	2 (9)
b. Equal participation	2 (20)	11 (48)
c. Reserved and passive	6 (60)	1 (4)
d. No information	2 (20)	9 (39)
4. Versatility in coitus		
a. Conservative (one or two positions only)	7 (70)	3 (13)
b. Experimental (many positions)	1 (10)	9 (39)
c. No information	2 (20)	11 (48)
D. Erotic zones		
1. Breasts		
a. Yes	7 (70)	12 (53)
b. No	2 (20)	7 (30)
c. No information	1 (10)	4 (17)
2. Clitoris (clitoris site)		
a. Yes	7 (70)	16 (70)
b. No	2 (20)	4 (17)
c. No information	1 (10)	3 (13)
3. Vulva and vagina		
a. Yes	8 (80)	16 (70)
b. No	0	4 (17)
c. No information	2 (20)	3 (13)
E. Erotic stimuli		
1. Smells and tastes		
a. Yes	2 (20)	4 (17)
b. No	6 (60)	11 (48)
c. No information	2 (20)	8 (35)
2. Visual		
a. Yes	3 (30)	16 (70)
b. No	5 (50)	3 (13)
c. No information	2 (20)	4 (17)
3. Narrative		
a. Yes	4 (40)	15 (66)
b. No	5 (50)	4 (17)
c. No information	1 (10)	4 (17)
4. Tactile		
a. Yes	8 (80)	16 (70)
b. No	0	3 (13)
c. No information	2 (20)	4 (17)

[a] One of the two was questionable.

Table III. Satisfaction with Female Sex Role and Cosmetic and Clothing Interests in 10 Patients with the Androgen Insensitivity Syndrome Contrasted with 15 Patients with the Early-Treated Adrenogenital Syndrome

Type of information	Frequency and percent (in parentheses) of total	
	Testicular feminization (androgen insensitive) group ($N = 10$)	Early-treated adrenogenital group ($N = 15$)
A. Sex role preference		
1. Content or prefers to be a female	9 (90)	7 (47)
2. Ambivalent	1 (10)	5 (33)
3. Desire to be a male	0	3 (20)
B. Rating of feminine appearance (patient's own estimate)		
1. Above average	1 (10)	[a]
2. Average	6 (60)	
3. Dissatisfied	2 (20)	
4. No information	1 (10)	
C. Preference of styles		
1. Dresses exclusively	7 (70)	1 (7)
2. Dresses plus slacks and shirts occasionally	2 (20)	5 (33)
3. Slacks and shirts with dresses only occasionally	0	9 (60)
4. Slacks and shirts with dresses excluded	1 (10)	0
D. Clothing, makeup, and jewelry		
1. Strong interest	8 (80)	2 (13)
2. Moderate interest	0	8 (54)
3. Little to no interest	2 (20)	5 (33)

[a] No information.

purposes in Table II, are the same as those already reported in Ehrhardt *et al.* (1968*b*). The 15 patients with the early-treated adrenogenital syndrome, used for contrast purposes in Table III, are those already reported in Ehrhardt *et al.* (1968*a*). The adrenogenital syndrome is genetically, gonadally, and hormonally almost the antithesis of the androgen insensitivity (testicular feminizing) syndrome.[7]

PROCEDURE

From each patient's case record, data from interviews and from the Draw-a-Person Test and the Guilford–Zimmerman Temperament Survey are utilized in this paper.

Each patient was interviewed individually by one or more of the authors. Each had been followed on a longitudinal basis for from 1 month to 13 years, the median and the mean both being 6 years. In the four cases of married patients, all three authors interviewed the husbands. Seven mothers and two fathers of patients were also interviewed.

[7] Patients with the adrenogenital syndrome have a normal female karyotype (46, XX) and two ovaries. They respond normally to circulating androgens, of which they are supplied with a superabundance from the adrenal cortex. For this reason they are masculinized at birth. If unregulated on cortisone therapy, which used to be routinely the case until cortisone treatment in this disorder was discovered in 1950, they develop, postnatally, a virilized physique.

The volume of information available varied from case to case, dependent on the availability of the patient and the frequency of endocrine and postsurgical follow-up appointments. In interviews, standard data schedules of topics were followed (Money and Primrose, 1969), but the interviews themselves were flexible. Sections of interviews were recorded in full or summarized with the patient on tape. From the transcripts, tabulated charts were made, under the same headings as those used in Tables II and III, summarizing the information pertinent to psychosexual orientation and to sexual behavior. Every entry on the charts was checked independently by two investigators.

The Draw-a-Person (DAP) Test was administered to all 10 patients. Two drawings were requested. The first instruction was simply to draw a person. When she had finished this, the patient was instructed to identify the figure by age and sex, and then to draw a figure of the opposite sex.

All 10 patients completed the GZTS, the Guilford–Zimmerman Temperament Survey (Guilford and Zimmerman, 1949). This survey is a 300-item questionnaire with a possible *yes*, *no*, or *?* response for each item. Of pertinence to the present study is the score on the masculinity–femininity (M-F) interest scale.

FINDINGS: INTERVIEW DATA

Sex and eroticism data abstracted from interviews are classified in Table II according to sexual practices, erotic dreams and fantasies *vis-à-vis* actual sexual experience, libido, erotic zones, and erotic stimuli. Table II also contains contrast data, already mentioned, obtained from a group of 23 hormonally masculinized women with the late-treated adrenogenital syndrome.

When, in what follows, statistical comparisons are drawn between the two groups of Table II, the Fisher–Yates Test of Significance, employing 2×2 contingency tables,[8] is the one used.

Sexual Practices

Table II shows that none of the 10 androgen-insensitive patients had ever experienced homosexual relations, whereas at least 13 percent (possibly 18 percent) of the adrenogenital patients had had bisexual experience. The two (20 percent) androgen-insensitive patients who reported no sexual experience were high school students. Of six adrenogenital patients having no experience in sexual relations, four had practiced masturbation. The much higher incidence of masturbation in the adrenogenital group over the androgen-insensitive group, in those patients having no interpersonal sexual experience, might be attributable in part to an androgen-heightened libido level in the adrenogenital patients.

Erotic Dreams and Fantasies *vis-à-vis* Actual Sexual Experience

Ten of the adrenogenital patients (44 percent), with or without actual bisexual

[8] Statistical analyses employed the tables of the *Geigy Scientific Tables*, 6th ed., pp. 109–123.

experience, reported having had dreams or fantasies of a homosexual type, and one other, for whom no information was available on sexual dreams, had had bisexual experience. Thus a total of 11 adrenogenital patients (48 percent) had either homosexual experience, dreams, or both. By contrast, none of the 10 androgen-insensitive patients evidenced homosexual experience or fantasy. This difference between the two diagnostic groups is statistically significant ($p \leqslant 0.01$).

Libido

In rating their own libido level, 20 percent of the androgen-insensitive patients *versus* 48 percent of the adrenogenital patients estimated their own sex drive as greater than most other women's. This difference is not statistically significant ($p > 0.10$). The adrenogenital women made their estimates at a time when they had already been normalized on cortisone therapy. Otherwise, there would undoubtedly have been a statistical difference between the two groups, since several adrenogenital women spontaneously remarked that, before cortisone therapy, their sexual drive had been stronger, and even too strong.

The data on climax experiences are roughly comparable for the two diagnostic groups. The category "most of the time" under "climax experiences" (Table II) implies that it was the exception when the patient did not reach orgasm.

A more reserved, passive, and receptive role in heterosexual relations is traditionally the female stereotype, in contrast with the traditionally more assertive, active, and initiating role of the masculine stereotype. Among the adrenogenital patients 57 percent were predominantly initiating or equal participants *versus* 20 percent of the androgen-insensitive patients. This difference does not reach statistical significance ($p > 0.05$), but only because of the relatively high frequency of no information in this category. When patients falling into the "no information" group are eliminated, then the percentages change to 93 and 25 percent, respectively.[9]

On the criterion of versatility in coitus, if "no information" patients are again excluded, then 75 percent of adrenogenital women *versus* 12 percent of androgen-insensitive women experimented with a variety of positions in coitus.

Erotic Zones

The data of Table II indicate that similar proportions of androgen-insensitive and adrenogenital women estimated themselves as being erotically responsive in the breasts, clitoral area, and vulva or vagina.

Erotic Stimuli

Smells and tastes as erotic stimuli were rather unimportant in the sexual arousal pattern for both groups of patients, whereas tactile stimuli, including kissing and caressing, were important.

[9] This same procedure has been adopted elsewhere in what follows and accounts for what might appear as a discrepancy between percentages given in the text and numerals given in the tables.

The two groups differed with respect to sexual arousal from visual stimuli. Among patients reporting on the variable, 84 percent of the adrenogenital patients *versus* 37 percent of the androgen-insensitive ones gave positive replies. The difference between the two patient groups is statistically significant ($p \leqslant 0.05$). Of the three androgen-insensitive patients who reported visual stimuli as sexually arousing, two were the two patients who had estimated their libido as higher than that of most other women.

There were 44 percent of the androgen-insensitive patients reporting (four patients, all married) who obtained sexual or intensely romantic arousal from narrative stimuli, such as love scenes in novels, *versus* 79 percent of the adrenogenital patients. The difference between the two patient groups is not statistically significant ($p > 0.05$).

The content of the arousal, a longing that was equally sentimental and sexual for the loved one alone, and not satisfiable in his absence, was conventionally female rather than male for all the androgen-insensitive patients who reported erotic arousal from narrative or visual material. The content of the arousal for the majority of the adrenogenital patients was also conventionally female, following the pattern described above. However, some of the adrenogenital patients, who had had bisexual experience, reported arousal more conventionally male, the arousal having a strong genitopelvic component with little sentimental component and very possibly leading to masturbation in the absence of the partner.

Satisfaction with Female Sex Role

Psychosexual data abstracted from interviews are classified in Table III according to sex role preference and patient's own rating of feminine appearance. By way of contrast, the data of Table III include also those obtained from 15 early-treated girls with the adrenogenital syndrome. The majority (90 percent) of androgen-insensitive patients clearly preferred and enjoyed being females *versus* 47 percent of the adrenogenital patients. This difference is significant ($p \leqslant 0.05$). Only one androgen-insensitive patient, the same patient who declined to take regular estrogen replacement treatment (needed because of gonadectomy in early childhood), was ambivalent—ambivalent about being adult rather than simply female. She was influenced at least in part by her own estimation of her father's antagonism toward her. She wanted to grow strong and unfeminine so that she could one day retaliate, when he hit her, by engaging him in combat. Both parents had been extremely traumatized by the way in which the discovery of their daughter's condition was transmitted to them.

Two patients in the androgen-insensitivity group were dissatisfied with their personal appearance. It might seem ironic that one of the two dissatisfied patients was a professional model, until one knows that her dissatisfaction, amounting to a phobic anxiety, lay in the symptom of lack of axillary and pubic hair. The patient could not tolerate her incompleteness as a woman, in the early years of her adulthood, although she subsequently became more reconciled to the deficit. The second dissatisfied patient was the girl dissatisfied not so much with her appearance as with what it would become if she took estrogen and grew breasts.

Cosmetic and Clothing Interests

Table III also presents findings on cosmetic and clothing interests. In regard to clothing style preference, only one androgen-insensitive patient, the one who rejected estrogen replacement therapy, preferred slacks and shirts to the point where she refused to wear skirts and dresses. Of the adrenogenital patients, 60 percent clearly preferred slacks, shorts, and shirts to dresses. This difference between diagnostic groups in preference for masculine-derived styles is statistically significant ($p \leqslant 0.025$).

The majority (80 percent) of androgen-insensitive patients showed a strong interest in clothing fashions, makeup, stylish hairdos, and jewelry. The adrenogenital patients, as a group, did not show as strong an interest in these areas.

FINDINGS: TEST DATA

Draw-a-Person (DAP) Test

Although the percentages vary with the age of the subject population tested, approximately 85–95 percent of normal males draw their own sex first, while about 50–70 percent of normal females draw their own sex first (for reviews, see Money and Wang, 1966; Swensen, 1968). On the DAP test, 80 percent of the androgen-insensitive patients drew a female first. For such a small sample, this proportion is compatible with the results of studies on unaffected females but is in sharp contrast to those on normal males.

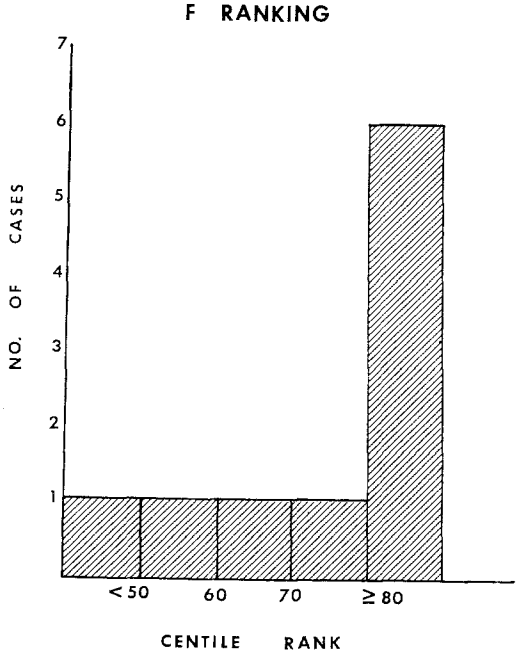

Fig. 1. Percentile ranking on femininity scale of Guilford–Zimmerman Temperament Survey for 10 patients with the androgen insensitivity syndrome. Note the high feminine ranking for the group.

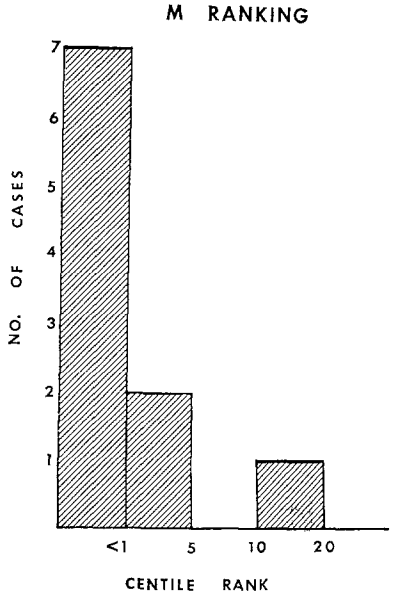

Fig. 2. Percentile ranking on masculine scale of Guilford–Zimmerman Temperament Survey for 10 patients with the androgen insensitivity syndrome. Note the low masculine ranking for the group.

M-F Scale, Guilford–Zimmerman Temperament Survey (GZTS)

The results on the M-F scale (masculinity–femininity interest scale) of the GZTS are shown in Figs. 1 and 2, according to the feminine and masculine rankings, respectively. Overall, the patients ranked very low on the masculine rating scale and high on the feminine scale, although the range on the feminine was broader than on the masculine. One point of special interest is that the girl who rejected estrogen therapy ranked below the 1st masculine percentile and above the 70th feminine percentile.

DISCUSSION

The two papers preceding this present one on the androgen insensitivity (testicular feminizing) syndrome demonstrated that women with this condition are typically womanly in their expectation of marriage, homemaking, and raising a family. They are distributed over the normal curve with respect to IQ, and do not show a masculine tendency for nonverbal to be higher than verbal abilities. The present paper extends the evidence of feminine psychosexual differentiation in the same group of patients to include self-reported erotic practices, dreams, zones, and stimuli; libido; sex-role preference; self-rating of feminine appearance; clothing and cosmetic preference; sex of first choice in human figure drawing; and femininity rating on a self-rating scale.

The evidence of the androgen insensitivity syndrome in the chromosomal (XY) male is therefore that the chromosomal constitution *per se* does not dictate psychosexual differentiation and does not directly determine masculinity or femininity of gender identity.

The genetic influence on psychosexual differentiation, one infers, must be indirect. The two most likely paths of influence are by way of the morphology and appearance

of the external genitalia, and the effect of hormones prenatally on the differentiation of the central nervous system. The brain region most likely to be implicated, if one judges on the basis of animal experimentation, is the hypothalamus.

The external genitalia in the androgen insensitivity syndrome differentiate as female, because the differentiating cells of the fetus are insensitive to androgen. In consequence, the child is assigned and reared as a female, and has the usual social experience of growing up as a female. One infers that the cells of the fetal brain, like those of the external genital anlagen, must have been resistant to the androgen secreted, in the manner typical for a male, by the fetal testes. In any case, there is, in subsequent behavior, no evidence of masculine traits, such as the tomboyism found in girls heavily androgenized in fetal life with the adrenogenital syndrome.

At puberty, cellular resistance to androgen again demonstrates itself in girls with the androgen insensitivity syndrome, as they go into a morphologically normal female puberty, except for amenorrhea (through absence of a uterine cavity) and infertility. The testes actually secrete normal amounts of androgen, but the body is capable of responding only to the estrogen normally produced in the male.

Pubertal feminization is consistent with the female gender identity which has typically differentiated during childhood in girls with the androgen insensitivity syndrome. Behavioral femininity, reinforced by the evidence of the body, continues with puberty and expands through adolescence.

Metaphorically speaking, one may say that the defeat of the XY sex chromosomal constitution has been complete, overpowered by the combined influence of sexual morphology, hormones, and rearing.

The differential effect of hormones *versus* rearing cannot be investigated in the androgen insensitivity syndrome, since no baby born with a normal-looking female vulva is assigned to be reared as a boy. There are, however, some instances of a *forme fruste* of the syndrome in which the baby is born with a phallic structure that looks like either an enlarged clitoris or a small, hypospadiac penis. In some instances (unpublished data) the child is assigned and reared as a boy, and differentiates a male gender identity, except for absence of masculine experience of genitopelvic functioning, orgasm, and parallel accompaniments of masculine cognitional eroticism.

These cases demonstrate that it is possible for psychosexual differentiation as a male to take place, even when not reinforced by the usual masculinizing hormonal effects prenatally and at puberty. Nonetheless, chromosomal genetics notwithstanding, it is far preferable that a person whose body can never achieve normal pubertal virilization should have been assigned, reared, and surgically corrected as a girl. When prenatal hormonal effects, surgical correction, rearing, and pubertal hormonal effects are well congruent with one another, and with gender identity, then one has achieved the optimal therapeutic synthesis. One has also maximized the individual's chance for happiness and fulfillment in life.

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