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Sexual orientation and medical history among Iranian people with Complete Androgen Insensitivity Syndrome and Congenital Adrenal Hyperplasia

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ABSTRACT

Objective: To report sexual orientation, relationship status and medical history of Iranian people with Differences of Sex Development (DSD) who were raised female.
Methods: Our participants consisted of nineteen 46,XY individuals with Complete Androgen Insensitivity Syndrome (CAIS) and eighteen 46,XX individuals with Congenital Adrenal Hyperplasia (CAH) who were raised as females and older than 13 years. As well as their relationship status and detailed medical history, an expert psychiatrist assessed their sexual orientation by a semi-structured psychiatric interview with them and, where applicable, their parents.
Results: Five percent of CAH participants and 42% of CAIS participants were in a relationship, which was significantly different. All CAH individuals had been diagnosed at birth; 89% of CAIS had been diagnosed after puberty and due to primary amenorrhea and 11% were diagnosed in childhood due to inguinal hernia. Genital reconstructive surgery had been performed in 100% of CAH participants and 37% of CAIS. Regarding sexual contact experiences and sexual fantasies (androphilic, gynephilic or both), no significant differences were found. However, CAH females had significantly more gynephilic dreams ($P = 0.045$).
Conclusion: This study, notable as one of the rare from a non-western culture, described sexual, medical and socioeconomic status of 46,XX CAH and 46,XY CAIS individuals living in Iran. Although broadly in line with previous findings from Western cultures, Iranian CAH individuals had fewer romantic relationships, but in contrast to previous studies their sexual orientation was only different from CAIS in the contents of sexual dreams.
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1. Introduction

Gender assignment in a newborn with ambiguous genitalia can be a clinical challenge. Traditionally the appearance of the external genitalia and factors such as maintaining the capacity for reproduction were regarded as important contributors to such decision making. However, due to the enormous increase in studies of the psychological, psychosexual[1] and quality of life outcomes in people with Differences of Sex Development (DSD), traditional approaches are being challenged. Considering the serious distresses that can result from the discordance

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[1] Psychosexual outcome is an umbrella term for three distinct concepts; **Gender Identity** (A person's concept of self as being male and masculine or female and feminine, or ambivalent, based in part on physical characteristics, parental responses, and psychological and social pressures), **Sexual Orientation** (an enduring pattern of romantic or sexual attraction (or a combination of these) to persons of the opposite sex or gender, the same sex or gender, or to both sexes or more than one gender) and **Gender Roles** (a set of societal norms dictating the types of behaviors which are generally considered acceptable, appropriate, or desirable for people based on their actual or perceived sex or sexuality) [39].

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between the felt gender identity and gender assigned at birth, some argue that the expected gender identity and sexual orientation in each specific DSD condition should be regarded as the most critical factors in gender allocation at birth [3].

Appropriate decision making at birth on gender assignment is particularly critical in countries such as Iran where a binary understanding of sexuality prevails and sexuality of citizens is the subject of much legislation. According to Islam, the official and governmental religion of Iran, men and women have different roles and in most cases are forcefully segregated both within families and in the social environment. This division starts at elementary school and is carried through to university. All forms of public transportation and most urban leisure spaces are also divided by gender [33]. Classical Islamic thought recognizes four genders among human beings: male, female, DSD/intersex (*khunsa*), and the effeminate male (*mukhannath*), however, in most of Islamic countries the latter two "recognized" genders are transitioned into one of the first two, as early as possible, to avoid any difficulties with religious regulations [42]. In addition to the issue of non-binary gender identities and sex segregation, none of the current official Islamic schools of thought tolerate any alternative to heterosexual sexual orientation[2] [15]. These factors make appropriate gender allocation to DSD newborns particularly critical in an Islamic culture, however, studies on the psychosexual outcomes of these individuals are lacking.

Congenital Adrenal Hyperplasia (CAH) and Complete Androgen Insensitivity Syndrome (CAIS), are the most frequently studied DSD condition. This is due in part to their increased prevalence compared to other DSDs [30] and also to their particular endocrinologic and genetic pathogenesis, which provides a unique opportunity to study the influence of prenatal androgens and sex chromosomes on gender and sexual orientation development [41].

In the classic form of CAH, which is the most common, 21-hydroxylase enzyme is deficient and cortisol production is impeded [13]. Consequently, the predecessor substrates such as adrenal androgens, progesterone, and 17-hydroxyprogesterone accumulate [23,35]. Elevated androgen level in female fetuses with classic CAH masculinizes their external genitalia so that in most cases surgical interventions after birth are performed. It should be mentioned, however, that surgical interventions in CAH for genital reduction is strongly questioned today from a human rights perspective [25]. This fetal hyperandrogenemia in 46,XX individuals with CAH can lead to masculinized brain organization and, consequently, at later stages of development, to masculinized gender-related behavior and cognitive function, including, albeit relatively uncommonly, gender identity [4]. Therefore, CAH females provide an opportunity to study the influence of prenatal testosterone on psychosexual development [13].

Studies on sexual orientation in CAH females suggest that these individuals are more likely to be homo- or bi-sexual [13]. One study of sexual orientation in 62 CAH females reported homo- or bisexuality in 19% of CAH compared to 2% of age-matched controls [7]. In a similar study evaluating 22 women with CAH, 13 (59%) had no physical involvement with females, eight (36%) had some involvement including kissing, fondling, petting and oral sex, and three of 22 (14%) had experienced genital to genital contact [19]. In another study, the sexual orientation of 143 women with CAH who were classified according to their clinical and molecular condition were compared to a group of non-CAH typical females who were sisters and female cousins of CAH participants using the Sexual Behavior Assessment Schedule (SEBAS). It was found that although most CAH women were heterosexual, the rates of bisexual and gynephilic orientation were increased above controls not only in women with classical CAH, but also in non-classic CAH women, and correlated with the degree of prenatal androgenization [25].

In contrast to CAH, 46,XY individuals with CAIS carry a mutation in an androgen receptor gene that renders them insensitive to typical levels of testosterone [28]. Thus, the genital phenotype of these individuals at birth is feminine and they are invariably assigned female gender roles [26]. As with external genitalia, all other aspects of sexual differentiation that are influenced by androgens develop in a typically female fashion in 46,XY CAIS individuals. CAIS may therefore provide additional insight into the influence of androgens on psychosexual development. Because of rarity of this condition, however, fewer studies have been conducted in CAIS than CAH.

In one of the first studies on CAIS, 22 individuals were recruited and, among other measures, their gender-related psychological characteristics (gender identity, sexual orientation, and gender role behavior in childhood and adulthood) and associated personality traits were evaluated [12]. All participants showed feminine childhood play behavior, gender identity, and personality characteristics, and authors concluded that psychological outcomes in women with CAIS are similar to those in other women. Twelve years later in a review of studies on the effect of androgen on psychosexual development, the same authors concluded that individuals with CAIS are almost always heterosexual [13]. In another study on a sample of German adults with XY DSD, CAIS individuals mainly reported heterosexual partners. In contrast, Kohler et al. reported that 46,XY female-raised individuals with Partial Androgen Insensitivity Syndrome (PAIS) were more likely to have sexual partners of the same and both sexes, thus further supporting the notion that androgen effects sexual orientation [21]. Interestingly, functional neuroimaging of 46,XY women with CAIS, typical (46,XX) women, and typical (46,XY) men has shown that brain responses of CAIS participants to various tasks (in which responses are usually different between men and women) are similar to typical women, and significantly different to that of typical males [8,38]. In spite of the findings of the latter investigations, however, there are recent studies reporting that psychosexual outcome in people with CAIS is more varied than previously believed. These studies show that androgen is not the only factor influencing the psychosexual development; epigenetics [29] and genetic factors such as fraternal birth order, genetic imprinting or sex chromosome Y effects, as well as non-biological elements such as gender allocation, sex of rearing and cultural features, may also play an important role [2,3,6,24,36].

Although some of the participants of the above studies may have had non-Western origins, the studies themselves were conducted in a Western, educated, industrialized, rich and democratic (WEIRD) [9] social context, in which gender discrimination, sex-based prejudices, stereotypes and stigmas have significantly reduced in the social atmosphere in recent decades. Studies on individuals with DSD from non-WEIRD cultures are, however, very scarce if not non-existent. This may be due, in part, to the fact that sexual orientation data in traditional societies is less accessible and less likely to be regarded as a subject for inquiry.

The aim of this study is thus to describe the sexual orientation, relationship status and medical history of a cohort of CAH and CAIS individuals, who live in the female gender role since birth, in a non-WEIRD country where a binary understanding of sexuality and gender is pervasive both legally and culturally. We also tried to compare outcomes between CAH and CAIS participants to test the possible effect of testosterone. To our best knowledge, this is the first study investigating sexual orientation of two DSD conditions in a non-Western context.

2. Materials and methods

This study was conducted within the framework of a research project, "Human Sex Differences among female gender-assigned individuals with CAH, CAIS and 5-alpha Reductase Deficiency" at the Mashhad University of Medical Sciences (MUMS). The sampling process has been explained in detail elsewhere [20]. The project was initially designed to investigate the psychosexual outcome of people with diversity

[2] Throughout this article, sexual orientation is defined "as a pattern of romantic or sexual attraction (or a combination of these) to persons of the opposite sex or gender, the same sex or gender, or both sexes or more than one gender. These attractions are generally subsumed under heterosexuality, homosexuality, and bisexuality" [1].

in their sex development in the context of a non-Western culture in which sexuality is exclusively understood as a binary concept. Research was focused on women because in an Islamic country such as Iran the sexuality of women is subject to more cultural and legal strictures than that of men, and thus their psychosexual development is more likely to be affected. CAH and CAIS conditions were chosen because their particular underlying endocrinologic physiopathology provides a unique opportunity to evaluate the effect of prenatal androgens on psychosexual development.

2.1. Participants

We analyzed the database of all hospitals affiliated with MUMS, the second largest medical center in Iran. We used following exclusion criteria: A) Patients under age 13[3], B) Patients with intellectual disabilities (indicated from the medical charts), C) Patients with any symptom suggestive of an underlying condition other than or in addition to CAH and CAIS, D) Patients with sex chromosome DSD with or without mosaicism, E) Male gender assignment at birth, F) Lacking chromosome karyotype in medical records, G) Lacking hormonal profile in their medical records and H) Lacking a valid address or contact details. Finally, n = 18 individuals with CAH and n = 19 individuals with CAIS participated in our study. The procedure of the study and the confidentiality of their identity information were explained to the patients, who all consented to join the study. After obtaining written informed consents from participants (and their parents if the participant was under the legal age), they were visited by a team of endocrinologists from MUMS able to distinguish CAIS and CAH from their differential diagnoses. These visits took place at the Endocrinology and Metabolism Research Center of MUMS and in their weekly session which is held to discuss endocrine equivocal cases. The final diagnosis of all patients was based on hormonal profile, karyotyping and gene sequencing of *AR* gene.

2.2. Measures

2.2.1. Demographics

Demographic data including age and socioeconomic (SES) status were measured using a questionnaire. The criteria that were used for measuring SES was educational level (highest level attained), head of the household's educational level, marital status, house ownership and number of people living together in the current house [14].

2.2.2. Medical and sexual history

Past medical and surgical histories of participants were obtained from their medical records. Some information such as the Prader stage of genitalia at birth was not always documented in the medical charts, and thus, not all such results were available to us. Where possible, therefore, parents of participants were also interviewed to gather lacking data. Among other things, parents were specifically asked about the amount of virilization at birth (particularly for CAH people), hormone therapy and surgical history. Prader stage 0 was considered as no virilization, Prader stages 1, 2 and 3 were considered as medium virilization and stage 4 and 5 were considered as high virilization.

A medical and sexual and social history was obtained in a semi-structured interview by a female psychiatrist with expertise in DSD and gender dysphoria. The structure of the interview was developed by the authors. The gender identity of participants was assessed using a DSM-IV based Gender Identity Disorder interview with a psychiatrist. To measure the sexual orientation of our patients, we did not use a questionnaire. The rationale was that due to the cultural sensitivity of the issue, the answers provided by patients would be more likely to be honest and accurate in an interview in which rapport with the person could be obtained before asking about their sexuality. The following three issues were discussed with all the patients: *1) Have you ever had any sexual contact/relationship with a man or with a woman? 2) Have you ever fantasized about having sex with a man or with a woman? 3) Have you ever dreamt about having sex with a man or with a woman?*

[3] The legally approved age for female marriage is 13 years of old according to the Iran's national law. However, based on Islamic law girls can marry as soon as they become 9 years old.

2.3. Procedure and ethics

Consenting participants (18 CAH and 19 CAIS patients) were invited to the Psychiatric and Behavioral Sciences Research Center lab at Ibn-e-Sina Psychiatric hospital, for psychological evaluations. Participants and their parents (if they were accompanied) were interviewed separately with the psychiatrist to obtain a detailed life and medical history. Parents were specifically asked about the amount of virilization at birth (for CAH group), hormone therapy and surgical history. The psychiatrist who interviewed the participants was unaware of the medical diagnosis of the patients. All patients were interviewed in the same location and with the same psychiatrist. Each interview lasted approximately 60 min.

The study conformed to the 1995 Declaration of Helsinki and was approved by the Ethics Committee of Mashhad University of Medical Sciences.

2.4. Statistical analysis

We used descriptive statistics to report the demographic characteristics, medical history, and sexual history outcome among CAH and CAIS groups. We used a Student's *t*-test for age comparison, and chi-square test for education, SES, and sexual history outcome between CAIS and CAH groups. We also used chi-square to compare the various parameters of sexual histories among groups with different amounts of virilization. The threshold for significance in these comparisons was considered to be a *P* value of 0.05. All analyses were conducted using SPSS version 16.0.1 for Windows.

3. Results

3.1. Demographics

Overall, 18 patients with CAH with a mean age of 17.11 (SD = 3.34, median = 16.00, range 14 to 26), and 19 patients with CAIS with a mean age of 20.53 (SD = 4.56, median = 21.00, range 14–28) participated in our study. CAH patients were significantly younger than CAIS, $t(35) = -2.59$, $P = 0.014$. Last obtained educational degree and SES did not differ significantly between CAH and CAIS groups ($P = 0.916$ and $P = 0.258$, respectively; Table 1).

Table 1
The socioeconomic and educational status of participants.

		CAH[a] (n = 18)	CAIS[b] (n = 19)
Age	M (SD)	17.1 (3.34)	20.5 (4.56)
	Range	14.0–26.0	14.0–28.0
	Median	16.0	21.0
Last obtained degree n (%)			
	Drop out	1 (6)	0 (0)
	High school	9 (50)	7 (37)
	Diploma	6 (33)	4 (21)
	Bachelor	1 (6)	5 (26)
	Master	1 (6)	3 (16)
Socioeconomic status n (%)			
	Low	1 (6)	2 (11)
	Mediate to low	4 (22)	5 (26)
	Mediate	7 (39)	8 (42)
	Mediate to high	4 (22)	3 (16)
	High	2 (11)	1 (5)

[a] CAH, Congenital Adrenal Hyperplasia.
[b] CAIS, Complete Androgen Insensitivity Syndrome.

3.2. Past medical history

In all cases of CAIS none was suspected to have DSD immediately after birth; 89% presented with primary amenorrhea at puberty and 11% (two out of 19) with inguinal hernia in childhood which were found to be testes. Along with hormone treatment, all the CAIS patients were gonadectomized and 37% had vaginoplasty. In contrast, all of our CAH patients had ambiguous genitalia at birth examinations (61% with medium virilization and 39% with high virilization), and were immediately diagnosed based on their hormonal profile. Hormone therapy was initiated immediately after diagnosis. Detailed information for each CAH and CAIS patient can be found in Tables 2 and 3, respectively.

All CAH patients had been taking glucocorticoid replacement therapy since diagnosis. However, the therapeutic management of these patients was modulated according to clinical response, and thus dosage varied not only between patients but also over the life time of each patient. The reconstructive surgery in all CAH cases consisted of clitoroplasty which was sometimes – in 7 out 18 (39%) – accompanied by labioplasty. None of the patients had undergone adrenalectomy or gonadectomy surgery. In the CAIS group, 100% of participants had been gonadectomized as a preventive treatment for gonadal malignancies and 37% had reconstructive surgery in form of vaginoplasty.

The mean age of patients at the time of their surgery was 4.39 (SD = 2.03, Range = 14–26, Median = 16) for the CAH group and 16.11 (SD = 1.19, Range = 14–28, Median = 21) for CAIS (t (35) = −21.5, $P < 0.001$). According to participants with CAH and their parents, all the surgeries were performed without seeking patients' consent because of their young age. However, none of the CAH patients had any complaint about their surgeries. In contrast, CAIS participants had all been referred to a pediatric psychiatrist at the time of diagnosis to make a decision about their treatment. All of them asserted that they had consented to their surgeries.

3.3. Sexual orientation outcome

All of participants were living with a female gender at the time of participation in the study. Tables 4 and 5 show various aspects of CAH and CAIS sexuality, respectively as reported in their interview.

CAH and CAIS participants were compared for their sexual contacts, sexual fantasies and sexual dreams. Regarding sexual contact, three out of 18 (17%) patients with CAH reported contact with men and a further two (12%) had experienced contact with women. In the CAIS group eight out of 19 (42%) participants reported sexual contact with men, of which one (5% of all CAIS) also reported sexual contact with women. Seven of the CAH participants (39%) and 13 of the CAIS participants (68%) reported androphilic fantasies while one of the CAH individuals (5%) and none of the CAIS patients reported to have had gynephilic fantasies. Two patients from both patient groups (11% for each) reported to have experienced both gynephilic and androphilic fantasies. Considering the contents of dreams, 3 of 18 people with CAH (17%) and 10 of 19 CAIS cases (52%) reported exclusively androphilic dreams while a further three CAH patients (17%) and no CAIS patients reported exclusively gynephilic dreams. Five of the CAH participants (28%) and only two CAIS participants (11%) reported to have experienced both gynephilic and androphilic dreams. A significant difference was observed only in sexual dreams, in which CAH patients reported more gynephilic content (χ^2 (3) = 8.034, $P = 0.045$). Additionally, when relationship status was compared, a significantly higher proportion of CAH participants were found to be single than in the CAIS group (χ^2 (3) = 10.135, $P = 0.017$). The mean age of CAIS patients at the time of their marriage was 17.12 (SD = 2.16, min = 15, max = 22). There was no significant difference between the marriage age of CAIS individuals to the age of CAH participants at the time of the study.

Table 6 describes the sexual history of individuals in each group based on whether they are younger or older than the median age of

Table 2
Medical description in CAH individuals.

Participant no.	Age at study (years)	Virilisation extent at birth	Hormone treatment	Age at initiation of hormone therapy (years)	Surgeries performed for the participant to the date of study	Age at first surgery (years)
1	14	Medium	Hydrocortisone + fludrocortisone	At birth	Clitoroplasty	2
2	14	Medium	Hydrocortisone	At birth	Clitoroplasty	10
3	14	Medium	Hydrocortisone	At birth	Clitoroplasty	5
4	14	Medium	Hydrocortisone + fludrocortisone	At birth	Clitoroplasty	4
5	15	High	Prednisone + fludrocortisone	At birth	Clitoroplasty + labioplasty	2
6	16	Medium	Prednisone + fludrocortisone	At birth	Clitoroplasty	6
7	16	Medium	Prednisone + fludrocortisone	At birth	Clitoroplasty	6
8	16	High	Prednisone + fludrocortisone	At birth	Clitoroplasty + labioplasty	5
9	16	Medium	Hydrocortisone + fludrocortisone	At birth	Clitoroplasty	3
10	16	High	Prednisone + fludrocortisone	At birth	Clitoroplasty + labioplasty	4
11	16	Medium	Prednisone + fludrocortisone	At birth	Clitoroplasty	2
12	17	Medium	Hydrocortisone + fludrocortisone	At birth	Clitoroplasty	4
13	17	Medium	Prednisone + fludrocortisone	At birth	Clitoroplasty	6
14	18	High	Dexamethasone + fludrocortisone	At birth	Clitoroplasty + labioplasty	4
15	18	High	Dexamethasone + fludrocortisone	At birth	Clitoroplasty + labioplasty	2
16	22	High	Prednisone + fludrocortisone	At birth	Clitoroplasty + labioplasty	5
17	23	High	Prednisone + fludrocortisone	At birth	Clitoroplasty + labioplasty	3
18	26	Medium	Dexamethasone + fludrocortisone	At birth	Clitoroplasty	6

Table 3
Medical description in CAIS individuals.

Participant no.	Age at study (years)	Virilisation extent at puberty	Hormone treatment	Age at initiation of hormone therapy (years)	Surgeries performed for the participant to the date of study	Age at first surgery (years)
1	14	None	Estrogen	14	Gonadectomy	14
2	15	None	Estrogen	14	Gonadectomy	14
3	16	None	Estrogen + progesterone	15	Gonadectomy	15
4	16	None	Estrogen	16	Gonadectomy	15
5	16	None	Estrogen	16	Gonadectomy	15
6	17	None	Estrogen	15	Gonadectomy	15
7	17	None	Estrogen	16	Gonadectomy	16
8	18	None	Estrogen	17	Gonadectomy	17
9	19	None	Estrogen + progesterone	17	Gonadectomy + vaginoplasty	16
10	21	None	Estrogen + progesterone	17	Gonadectomy + vaginoplasty	17
11	21	None	Estrogen	18	Gonadectomy	17
12	22	None	Estrogen + progesterone	18	Gonadectomy + vaginoplasty	18
13	22	None	Estrogen + progesterone	20	Gonadectomy	16
14	23	None	Estrogen	18	Gonadectomy + vaginoplasty	17
15	24	None	Estrogen	17	Gonadectomy	17
16	26	None	Estrogen	15	Gonadectomy + vaginoplasty	16
17	27	None	Estrogen	18	Gonadectomy + vaginoplasty	18
18	28	None	Estrogen	18	Gonadectomy + vaginoplasty	17
19	28	None	Estrogen	16	Gonadectomy	16

their group. While CAH patients who were older than median age had a higher rate of sexual contacts rather than those who were younger ($\chi^2(2) = 10.879$, $P = 0.004$), no differences were found in their sexual fantasies and dreams. In the CAIS group, no difference between old and young participants was found.

Finally, sexual histories of all participants were compared based on the amount of their genital virilization at birth. No significant difference was found among groups with various stages of (none, medium and high) virilization.

4. Discussion

Elevated prenatal androgens are proposed to masculinize the psychosexual development of CAH women; making them less androphilic and more gynephilic. In contrast, it has been suggested that in CAIS women, who are insensitive to androgens, androphilia is more likely. This study reports on clinical features and sexual orientation of a sample of 46,XX CAH/46,XY CAIS individuals who were all raised as female. We tried to infer sexual orientation based on three variables: sexual experiences, dreams, and fantasies. We found that our CAH women reported a higher rate of gynephilic experiences, dreams, and fantasies than the CAIS group. However, perhaps due to the small sample size, comparisons between the two groups revealed a significant difference only in frequency of gynephilic dreams.

Published literature on sexual orientation of girls and women with CAH implies a lower probability of androphilia compared to a non-clinical female control group [10,11]. Almost 5% of women in general population have gynephilic orientation, but among those with CAH, this proportion increases to 30% [34]. Among CAH patients in our study, two (11%) reported exclusively gynephilic sexual experiences, three

Table 4
Psychosexual characteristics in CAH Individuals.

Participants number	Current age	Lives as	Relationship status	Have you ever had any form of sexual contact with		Have you ever fantasized about having any form of sexual contact with		Have you ever dreamt about having any form of sexual contact with	
				Men	Women	Men	Women	Men	Women
1	14	Female	Single	–	–	–	–	–	–
2	14	Female	Single	–	–	–	–	–	–
3	14	Female	Single	–	–	Yes	–	–	–
4	14	Female	Single	–	–	–	–	–	–
5	15	Female	Single	–	–	–	–	–	–
6	16	Female	Single	–	–	Yes	–	Yes	Yes
7	16	Female	Single	–	–	Yes	–	Yes	–
8	16	Female	Single	–	–	–	–	–	–
9	16	Female	Single	–	–	Yes	–	–	–
10	16	Female	Single	–	–	–	–	–	Yes
11	16	Female	Single	–	–	–	–	Yes	Yes
12	17	Female	Single	Yes	–	Yes	–	Yes	Yes
13	17	Female	Single	–	Yes	–	Yes	–	Yes
14	18	Female	Single	–	Yes	Yes	Yes	Yes	Yes
15*	18	Female	Single	–	–	–	–	Yes	–
16*	22	Female	Married	Yes	–	–	Yes	–	Yes
17*	23	Female	Single	–	–	Yes	–	Yes	Yes
18	26	Female	Single	Yes	–	Yes	–	Yes	–

* Congenital Adrenal Hyperplasia.

Table 5
Psychosexual characteristics in CAIS Individuals.

Participants number	Current age	Lives as	Relationship status	Have you ever had any form of sexual contact with		Have you ever fantasized about having any form of sexual contact with		Have you ever dreamt about having any form of sexual contact with	
				Men	Women	Men	Women	Men	Women
1	14	Female	Single	–	–	Yes	–	Yes	–
2	15	Female	Single	–	–	–	–	–	–
3	16	Female	Single	–	–	Yes	–	–	–
4	16	Female	Single	–	–	Yes	–	Yes	–
5	16	Female	Engaged	Yes	–	Yes	–	Yes	–
6	17	Female	Married	–	–	–	–	–	–
7	17	Female	Engaged	Yes	–	Yes	–	Yes	–
8	18	Female	Single	–	–	Yes	–	–	–
9	19	Female	Married	Yes	–	Yes	–	Yes	–
10	21	Female	Single	–	–	Yes	–	Yes	–
11	21	Female	Married	Yes	–	Yes	Yes	Yes	Yes
12	22	Female	Single	–	–	–	–	–	–
13	22	Female	Single	–	–	–	–	–	–
14	23	Female	Married	Yes	–	Yes	–	–	–
15	24	Female	Single	–	–	Yes	–	Yes	–
16	26	Female	Engaged	Yes	–	Yes	–	Yes	–
17	27	Female	Married	Yes	–	Yes	–	Yes	–
18	28	Female	Single	Yes	Yes	Yes	Yes	Yes	Yes
19	28	Female	Single	–	–	Yes	–	Yes	–

(17%) reported gynephilic fantasies and nearly half (8 out of 18) of them have dreamt forms of sexual contact with women. Since, to our best knowledge, this is the first study of prevalence of sexual orientation variations in Iranian patients, we had to compare these proportions to other datasets which were invariably obtained from cohorts in Western countries. The rate of gynephilia in our cohort is higher than would be expected in the general population, but lower than the rate of gynephilia in CAH women in other studies from Western countries. This may reflect the cultural stigma and legal consequences of non-heterosexual orientation in Iran that make it difficult for participants to openly express their genuine sexual feelings. It may also be related to the relatively young age of our participants; it has been shown that CAH females are more likely to show gynephilic orientation as they get older [5]. The suggested reason for the latter observation is that when adolescents grow older into the adulthood they are more likely to recognize their true sexual orientation [10,11]. In our study, we too found a significant higher prevalence of sexual contacts in those who were older than median age, but this was only true in the CAH group. Interestingly, however, even in the CAH group no significant difference was found in the rate of sexual fantasy and sexual dreams between older and younger participants. One may conclude that the change that occurs from adolescence to adulthood is not more accurate recognition of sexual orientation but only ability or inclination to express previously suppressed feelings. The fact that no difference was detected in any sexual aspects of young and old participants with CAIS suggests that change in sexual orientation from adolescence to adulthood may not be true for all kinds of DSD.

None of the CAH participants who were younger than 17 years of age reported experience of sexual contact. The fact that our CAH participants were significantly younger than those with CAIS should be considered when interpreting the latter observation, but it should also be noted that our analysis showed that the age at which our CAIS participants had married was not significantly different from the age of our CAH women at the time of the study. This argues against young age as the only reason for absence of sexual contact in our CAH girls. Additionally the role of social and cultural hostility toward same-sex experiences should be considered when interpreting these data; CAH patients with non-heterosexual tendencies may have fewer sexual contacts because their desired experience is not only socially unacceptable but also punishable in an Islamic traditional context.

Higher gynephilia or bisexuality in CAH women has been correlated with higher degrees of genital masculinization at birth or with more severe types of the disease [25]. However, we could not find any meaningful pattern in the sexual orientation of our patients according to degree of virilization of genitals at birth. This may be due to our limited sample size.

In our study, almost 70% of CAH patients reported no history of sexual contact. A potential explanation is that genital surgeries in female CAH individuals can be traumatizing and can produce anxieties and a subsequent reluctance to experience sexual contacts. A review article

Table 6
Description (number and percentage) of sexual history among CAIS and CAH Individuals.

N (%)		Relationship status			Sexual contacts				Sexual fantasy				Sexual dream			
		S	Ma	E	M	W	MW	None	M	W	MW	None	M	W	MW	None
CAH	Younger[a]	11 (100)	0	0	0	0	0	11 (100)	4 (36.4)	0	0	7 (63.6)	1 (9.1)	1 (9.1)	2 (18.2)	7 (63.6)
	Older[a]	6 (85.7)	1 (14.3)	0	3 (42.9)	2 (28.6)	0	2 (28.6)	3 (42.9)	2 (28.6)	1 (14.3)	1 (14.3)	2 (28.6)	2 (28.6)	3 (42.9)	0
	Comparison[b]	$P = 0.389$			$P = 0.004$				$P = 0.069$				$P = 0.062$			
CAIS	Younger	6 (54.5)	3 (27.3)	2 (18.2)	4 (36.4)	0	0	7 (63.6)	8 (72.7)	0	1 (9.1)	2 (18.2)	6 (54.5)	0	1 (9.1)	4 (36.4)
	Older	5 (62.5)	2 (25)	1 (12.5)	3 (37.5)	0	1 (12.5)	4 (50)	5 (62.5)	0	1 (12.5)	2 (25)	4 (50)	0	1 (12.5)	3 (37.5)
	Comparison[b]	$P = 0.926$			$P = 0.466$				$P = 0.894$				$P = 0.965$			

*S, single; Ma, married; E, engaged; M, men; W, women; MW, men and women; N, none; M, medium; H, high.
[a] Each endocrine group is separated based on its median age into those who are younger than median age and those who are older than the median age.
[b] Using chi-square, sexual features between younger and older participants in each endocrine condition were compared.

of all studies on 46,XY DSD patients (living in male or female gender) between 1974 and 2007 that had evaluated the sexual quality of life, showed those with a history of genital surgery identified themselves more with the non-heterosexual orientation and reported fear of sexual contacts more frequently compared with patients lacking this medical history [34]. All of our CAH patients had undergone genital surgery and this may have contributed to the low rate of sexual experiences. It has also been suggested that if these interventions become traumatizing, they can contribute to androphilic orientation by causing fear of penetration in patients with DSD [21]. Such fears of penetration and sexual contact, thus, need to be acknowledged in assessing the medical need for a genital surgery [22]. Other explanations include the younger age of our CAH participants and the effects of living in a traditional society where premarital sexual contacts are strongly prohibited [18,27]. This is in congruence with findings of a study on Chinese patients in which only 14% of individuals with DSD were sexually active [40].

In addition to differences in sexual history, the prevalence of romantic relationships was significantly lower in CAH than CAIS participants. This is, too, in line with previous studies. One study found that DSD patients who had not been influenced by androgens any time during their sexual development were more likely to be in love relationships [17]. Concordantly no significant difference was found between CAIS women and their female controls regarding their marital patterns [12]. On the other hand, the 46,XX patients with an experience of increased levels of androgen during gestation (mostly CAH patients) were less frequently in a love relationship [5]. It has also been suggested that CAH is associated with a reduction in sexual interest [10,11]. Generally, females with CAIS have typical female external genitalia and this can reduce anxieties and insecurities which can be seen in patients that are afflicted with other forms of DSD. Furthermore, as noted above, the observed decrease in sexual function in women with CAH may in part be accounted for by genital virilization at birth and unsatisfactory results of subsequent genital surgery [37].

This is the first study of sexual orientation outcome among a rather large sample of DSD individuals in Iran. The size of the CAIS group in our study was particularly large relative to the rarity of this disease. To assess sexual orientation, some studies have relied only on the sex of the sexual partners [10,11]. However, studies that evaluated erotic imagery (with or without gender of sexual partners) to investigate sexual orientation have reported higher rates of reduced heterosexuality [10,11]. This, along with the sensitivity of the issue in the traditional Iranian culture, convinced us to take a more holistic approach by applying a semi-structured interview to investigate sexual orientation. In addition, we deliberately used a female interviewer because sex is considered to be a sensitive topic in Iranian public life and we believed that our participants would feel less inhibited with a female psychiatrist. Previously, some investigators have attempted to use a sub-grouping system based on chromosomal sex, androgen experience and the sex of rearing: 46,XX DSD with virilization (such as females with Congenital Adrenal Hyperplasia), 46,XY DSD with complete feminization (Pure Gonadal Dysgenesis and Complete Androgen Insensitivity Syndrome), 46,XY DSD with partial androgen effect raised as girls (Partial Androgen Insensitivity Syndrome, Androgen Biosynthesis Deficiencies), and 46,XY DSD with partial androgen effect raised as boys (Partial Androgen Insensitivity Syndrome) [31]. However, this strategy could impose inherent limitations by disregarding the possible influences of, for example, genetic imprinting, prenatal hormones and gender assignment on psychosexual development. To overcome this issue, we assessed and compared only two distinct types of DSDs: CAH and CAIS.

The number of CAIS patients in this study was large compared to many other studies; nevertheless a general limitation of this study was that the sample size remained too small for robust statistical comparison. Moreover, degrees of genital virilization at birth in CAH patients were obtained from medical files from different health centers which did not provide documented Prader stage. The absence of a controlled observation of genital virilization may mask any existing association between genital virilisation at birth and sexual orientation in these patients. Furthermore, our CAH patients were younger than CAIS cases and our study was cross-sectional in design. To fully appreciate the impact of age on sexual orientation outcomes in these conditions a longitudinal study would be required.

This study was unable to support the previous findings that CAH women are significantly more likely to have experienced homo- or bisexual contacts and fantasies, but the significant increase in gynephilic dreams in our Iranian CAH women suggest a trend toward a general increase in gynephilia that may prove to be significant in a larger cohort. We confirmed prior findings that indicate that XY CAIS females are mostly androphilic. The former observation is particularly important because it shows that despite social hostility toward any non-heterosexual orientation, some CAH women nonetheless develop gynephilic desires. Nevertheless, the fact that the rate of gynephilia was diminished in our sample compared to Western reports may indicate that cultural factors retain some degree of influence in forming sexual tendencies and behaviors. In addition, as a recent Indian study on psychological aspects of individuals with DSD reports social stigma and discrimination against patients as well as certain advantages in favor of one sex can strongly influence the gender development of children with DSD [16]. We were unable to confirm an association between degrees of genital virilization at birth and decreased heterosexuality in CAH females reported in some previous studies [25,32,37]. Precise information about sexual characteristics of individuals with DSDs is necessary for health professionals and practitioners to limit potential social distress that these persons can face concerning their sexual functioning. Therefore, further studies on sexual development of DSDs are needed, particularly from non-Western countries where the data is scarce.

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Conflict of interest statement

All authors have completed the Unified Competing Interest form at http://www.icmje.org/coi_disclosure.pdf. The authors have no competing interests to report. There has been no financial support for this work that could have influenced its outcome.

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