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ORIGINAL PAPER

Gender Development in Indonesian Children, Adolescents, and Adults with Disorders of Sex Development

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Abstract In most Western countries, clinical management of disorders of sex development (DSD), including ambiguous genitalia, begins at diagnosis soon after birth. For many Indonesian patients born with ambiguous genitalia, limited medical treatment is available. Consequently, affected individuals are raised with ambiguous genitalia and atypical secondary sex characteristics. We investigated gender identity and gender role behavior in 118 Indonesian subjects (77 males, 41 females) with different types of DSD in comparison with 118 healthy controls matched for gender, age, and residential setting (rural, suburban, or urban). In Study 1, we report on methodological aspects of the investigation, including scale adaptation, pilot testing, and determining reliability and validity of measures. In Study 2, we report on gender development in 60 children (42 boys, 18 girls), 24 adolescents (15 boys, 9 girls), and 34 adults (19 men, 15 women) with DSD. The majority of participants with DSD never received any medical or surgical treatment prior to this study. We observed a gender change in all age groups, with the greatest incidence in adults. Among patients who changed, most changed from female to male, possessed a 46,XY karyotype, and had experienced significant masculinization during life. Gender identity confusion and cross-gender behavior was more frequently observed in children with DSD raised as girls compared to boys. Puberty and associated masculinization were related to gender problems in individuals with 46,XY DSD raised female. An integrated clinical and psychological follow-up on gender outcome is necessary prior to puberty and adulthood.

Keywords Gender identity · Gender role behavior · Disorders of sex development · Intersexuality · Indonesia

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Introduction

Disorders of sex development (DSD) are characterized by, among other things, an atypical development of the anatomical sex (Hughes, Houk, Ahmed, & Lee, 2006). Many individuals with DSD are identified at birth or in childhood because they have ambiguous or atypically developed genitalia. In Western countries, children with ambiguous genitalia or adolescents with an atypical puberty will enter medical care for a diagnostic work up, followed by a medical treatment plan.

In most children with a 46,XX karyotype, genital ambiguity is caused by an excess of prenatal androgen production, whereas in most children with the 46,XY karyotype, genital ambiguity is due to insufficient androgen production or action. Significant exposure to androgens during critical periods of fetal development is linked to masculinization and defeminization of human behavior (Cohen-Bendahan, van de Beek, & Berenbaum, 2005; Hines, 2011a, b; Jürgensen, Hiort, Holterhus, & Thyen, 2007). This action of prenatal androgens on human behavior is in line with findings in experimental studies in non-human animal species (Wallen, 2005). Social influences play less of a role in influencing certain behaviors; for example, parenting style has less influence on masculinized behavior (Pasterski et al., 2005). Girls with excessive androgen exposure due to congenital adrenal

hyperplasia (CAH) display masculinized behavior (Berenbaum, 1999; Berenbaum, Duck, & Bryk, 2000; Hines, Brook, & Conway, 2004; Nordenström, Servin, Bohlin, Larsson, & Wedell, 2002; Slijper, 1984), and these findings have been replicated in young Indian patients (Ammini et al., 2002; Gupta, Bhardwaj, Sharma, Ammini, & Gupta, 2010). No other investigations of gender in patients with DSD have previously been conducted in other Asian countries.

The large majority of women with CAH identify themselves as women; however, gender dysphoria and a desire to live as male is more frequently seen in this group compared to the overall population of women (Dessens, Slijper, & Drop, 2005). Gender dysphoria and gender change also are seen in patients with 46,XY DSD due to partial androgen insensitivity syndrome (PAIS) and in patients with disorders in the biosynthesis of androgens (Mazur, 2005, Cohen-Kettenis, 2005). Among females with complete androgen insensitivity syndrome (CAIS), gender dysphoria and gender change are less common (Hines, Ahmed, & Hughes, 2003; Mazur, 2005; T'Sjoen et al., 2011).

In developing countries, physicians see patients who seek medical services later in life (Warne & Raza, 2008). Many patients with DSD and their parents will consult with a physician only after psychological or social problems with genital or body ambiguity have intensified. In Indonesia, medical management for patients with DSD is characterized by limitations in health resources and by sociocultural and legal barriers that restrict choices for affected individuals. Every Indonesian newborn must be assigned a female or male gender and be registered in the local civil registration office within 60 days in order to obtain a birth certificate. A birth certificate is compulsory for school entry and for obtaining a diploma, health insurance, and ID card. Delayed birth registration or change of gender in the birth registry requires a legal procedure in an Indonesian state court. In cases of gender change, either due to DSD or gender dysphoria (American Psychiatric Association, 2013), the court requests a medical review before reaching a decision about the change.

In 2010, the Indonesian Ulama Council (MUI) released its fatwa—a religious edict—to ban sex reassignment surgery for transsexual individuals. According to this fatwa, gender reassignment due to DSD is permitted within Islam sharia. Even if the sex reassignment procedures have not been carried out completely and the sex change has not yet been legalized by the court, the Islamic law regards the person as reassigned. The MUI also endorsed other parties (e.g., local parliaments, the Indonesian Medical Association, and the courts) to integrate this fatwa into the new regulation on gender reassignment (Haryadi, 2010). The MUI also released a formal letter supporting a child with 46,XY karyotype and ambiguous genitalia diagnosed with DSD to undergo a female-to-male gender reassignment (Raharjo, 2013).

Parents of newborns with ambiguous genitalia often report difficulties with the birth registration of their child. For many parents, there is no professional help available and the sociocultural and legal barriers are substantial. Our team has been confronted by patients who suffered from emotional problems because they lived in an ambiguous body, experienced gender confusion or gender dysphoria or suffered from social stigmatization. These reported difficulties form the basis of the current study.

Specifically, we assessed gender identity and gender role behavior in children, adolescents, and adults with DSD whose first presentation for medical assistance was delayed, and compare these observations to results from studies performed in Western countries.

Study 1: Scale Adaptation and Pilot Study for Gender Assessment

Prior to this study, there were no measures available in the local language (*Bahasa Indonesia*) to assess gender. Therefore, we used measures developed in Western countries that had been applied to assess gender in patients with DSD. Although translated and adapted measures differ from the original ones as adaptation to another language or culture may impact on item variance and factor structure and hence affects generalization, we decided to translate available questionnaires as, to some extent, comparison of findings in Indonesian, North American and European patients would be possible. The study protocol was approved by the board of the ethics committee at the Faculty of Medicine, Diponegoro University, Semarang, Indonesia.

Method

Scale adaptation was conducted through three stages: (1) translation into the local language (*Bahasa Indonesia*) by a certified translator; (2) review of the translations by a panel of researchers (AE, AD) and an anthropologist who had expertise in gender studies in Indonesian culture and understands both English and Indonesian languages, and (3) a pilot study involving controls and patients with DSD to evaluate the applicability and feasibility of using these measures. To assess reliability and validity of measures applied in children and adolescents, data were obtained from participants and matched control samples. To assess reliability and validity of measures applied in adults, we conducted a web-based survey.

Participants and Procedure

Pilot Study Participants

Thirty-six subjects, aged 6–25 years, raised male or female, were recruited for the pilot study group. This group comprised 6 patients (2 adults, 2 adolescents, 2 children) and 30 healthy controls (10 children, 10 adolescents, 10 adults). Half of the participants in each group were male. All 6 patients (3 with a 46,XX karyotype and 3 with a 46,XY karyotype) were living in

their assigned gender and resided in Semarang or nearby towns. The controls, who also resided in the Semarang area, were contacted through the local leaders (*Pak Lurah* or *Pak RT* for children) or schools/university (for adolescents and adults) and joined the study voluntarily. After the adult participants and parents of subjects under age 18 provided their consent, questionnaires were administered and they received a gift (stationery or towel) for their time.

Assessment of Reliability and Validity

To assess reliability and validity of the measures applied to the child and adolescent participants, we used data from 120 children (60 with DSD and 60 matched controls) and 48 adolescents (24 with DSD and 24 matched controls) who participated in Study 2. This group was composed of 84 boys and 36 girls (ages 6–11 years) and 30 adolescent boys and 18 adolescent girls (ages 12–17 years).

Web-Based Survey Participants

The web-based survey was set up in the local language (*Bahasa Indonesia*). During a 4-week period, invitations to participate in the survey were emailed to mailing lists of Indonesian communities (e.g., university students, university graduates, Indonesian professionals' networks). Recipients were encouraged to forward the email invitation to their adult contacts. Study information (e.g., the study aim, the principal investigator and affiliations, the estimated time needed to complete the self-report, and confidentiality assurance) was provided on the welcome page prior to participation. Visitors to the web-survey page could give informed consent by ticking off a box prior to completing the survey. Alternatively, they could decline participation. Participants were not obliged to answer all questions. In total, 377 healthy Indonesian adults aged 18–45 years old joined the web-based study and 316 (84 %) completed the Gender Questionnaire, while 254 (65 %) completed the Activities Questionnaire.

Measures

To assess gender identity and gender role behavior in children, we applied the Gender Identity Interview for Children (GIIC) (Zucker et al., 1993) and the Gender Identity Questionnaire for Children (GIQC) (Wallien et al., 2009). For adolescent and adult samples, we applied the Gender Questionnaire and the Activities Questionnaire (Hines et al., 2003, 2004).

Gender Identity Interview for Children (GIIC)

The GIIC measures children's cognitive and emotional understanding of his or her gender and the desire to be the other sex (Wallien et al., 2009; Zucker et al., 1993). This scale consists of 12 items with a 3-option response mode ranging from 0 to 2: sex-appropriate response (score 0), ambiguous or intermediate response (score 1), and cross-sex response (score 2). In this study, we added one question referred to liking or disliking the external genitalia with a 3-option response mode: male genital, neither male nor female genital, female genital. Higher scores indicate more gender confusion.

Gender Identity Questionnaire for Children (GIQC)

The GIQC is a standardized measure to assess a parent's evaluations of their child's gender role behavior and cross-gender role behavior (Johnson et al., 2004). This Likert rating scale consists of 12 items with a 5-option response mode (from 1 to 5) measuring frequencies of various play and playmate preferences (ranging from never to always) and 4 items assessing the desire to be the other sex and anatomic dysphoria. The higher the score, the more sex typical the gender role behavior is (more masculine behavior in boys or more feminine behavior in girls).

Gender Questionnaire

The Gender Questionnaire aims to assess an individual's core gender identity, gender role behavior, and sexual orientation (Hines et al., 2003). For this study, we only used eight items measuring gender identity and gender role behavior (Items 1–8). We added the following questions to assess subjects' wishes for a social gender role change (in the version for women): "I was treated as a man by people who knew me well and people who were unfamiliar to me," "I present myself as a man during work or leisure time," "I want medical treatment to change the appearance of my body into a man's body." Two reference periods were applied for each additional item: in the past 12 months and during lifetime. In the male version, similar questions were applied by replacing the term "man" with "woman". In total, six items were added for the purpose of this study; therefore, the Indonesian version of Gender Questionnaire comprised 14 items. We provided two versions (female and male version) to the participants based on the gender they lived in at participation in the study. Similar to the original English version, a seven-option response mode ranging from always (1) to never (7) was used and reversed scoring was applied in Items 5–14.

Activities Questionnaire

The 14-item Activities Questionnaire measures the recalled childhood preferences for playmate, toys, or activities (Hines et al., 2003). Following the original version, a five-option response mode ranging from almost always (1) to not at all (5) was applied to assess stereotypically masculine, feminine, or neutral preferences, except in one question assessing preference for playmate that ranged from always girls (1) to always boys (5). Similar to the original scale, reversed scoring was applied in negatively worded items; therefore, higher scores reflect greater preferences in sex-typical preferences.

Table 1 Results of principal component analysis (PCA) and reliability analysis of measures used in the study

Measures (number of items applied in this study)	N	Number of components (% of total variance explained)	Components and item distributions	Cronbach's alpha (α)
Gender Identity Interview for Children (9 items)	120	2 PC (71.7)	Cognitive gender confusion (Items 1, 2, 6, 8, 10)	0.78
		1 PC (56.8)	Affective gender confusion (Items 3, 5, 7, 11)	0.86
			Overall gender confusion (9 items)	0.88
Gender Identity Questionnaire for Children (10 items)	120	1 PC (37.3)	Child's gender role behavior (parent report)	0.88
Gender Questionnaire (14 items)	316	2 PC (74.9)	Gender identity and gender role behavior (Items 1–4)	0.88
			Cross-gender identity and cross-gender role behavior (Items 5–14)	0.96
Activities Questionnaire/recalled childhood preference (12 items)	254	3 PC (55.2)	Masculine type of preference (Items 1, 3, 4, 5, 7)	0.87
			Feminine type of preference (Items 2, 10, 12)	0.73
			Neutral type of preference (Items 6, 8, 9, 11)	0.38

Statistical Analysis

Construct validity was explored using principal component analysis (PCA) with varimax rotation method and Kaiser normalization. Factors with Eigenvalues greater than 1 and items with factor loadings greater than .40 were considered acceptable. Instrument reliability was evaluated as internal consistency, with Cronbach's alpha. Several models were tested; here, we report the optimal model for the Indonesian data.

Results

Reliability and Validity

During the pilot, we learned that participants were willing to discuss gender-related issues; the paper–pencil methods in administering the measures was only effective for educated participants who were also familiar with self-report methods. For participants who received limited education, were illiterate, or who were unfamiliar with self-reports, oral application was the best way to obtain information. Oral application ensured participants understood the questions before they gave an answer. The results of PCA and reliability analysis (Cronbach alpha for internal consistency) are shown in Table 1.

Gender Identity Interview for Children (GIIC)

During the pilot study, we detected three problematic items in the Indonesian translation of GIIC: Item 2 ("Are you a ….?"—a boy or a girl opposite to first response given), Item 4 ("Could you ever grow up to be …?"—a boy or a girl opposite to first response given), and Item 11 that assess personae in dreams (numbering according to the original scale). Item 2 and Item 4 triggered unexpected responses from children (laughing or upset) and/or parents (irritated or suspicious) which reduced children's or parents' enthusiasm to respond to further questions. Regarding Item 11, most of the children reported they never have had dreams or that they dreamed about (being) a popular cartoon personality broadcasted on the Indonesian television. This cartoon hero was neither male nor female. We considered the influence of a culture-bias in these items. After reviewing these problematic items, we decided not to apply Items 2 and 4, but applied Item 11 carefully to see the possibility of different findings reported from study participants. Consequently, the item numbering differed from the original English version (see Table 2 for items applied in this study).

During the study, we identified two items with a low response rate: Item 6 ("Is there anything you don't like about being a boy?"—question given to a boy) and Item 11 assessing child-reported dreams. Due to the large proportion of missing answers (55–98 %), we excluded these items from statistical analyses and subsequently performed PCA and internal consistency analysis on the nine remaining items. The PCA generated two PC solutions and 71.66 % of the total variance was explained. All items had a loading factor greater than 0.60. Table 2 shows that Items 1, 2, 6, 8, and 10 loaded to Factor 1, whereas Items 3, 5, 7, and 11 loaded to Factor 2 (item numbers according to the version applied in this study). Factor 1 reflects the cognitive component of gender confusion and Factor 2 mirrors the affective component of gender confusion. The Cronbach's alphas obtained for Factor 1 ($\alpha = 0.78$), Factor 2 ($\alpha = 0.86$), and all nine items together ($\alpha = 0.88$) indicated that the Indonesian version of GIIC has a good internal consistency.

The results we found for the Indonesian sample differ from previous studies (Wallien et al., 2009; Zucker et al., 1993). Although Zucker et al. and Wallien et al. also found a 2PC model in their U.S. and Dutch samples, their items loaded differently on each principal component. In order to enhance the comparability with their results, we additionally report the results sum scores of

Table 2 Mean, SD, and factor loadings of the Indonesian version of GIIC items

Items (boy version)	$M \pm SD$	Factor loadings (2 PC)		Factor loadings (1 PC)
		Factor 1[a]	Factor 2[b]	
1. Are you a boy or a girl?	0.02 ± 0.18	**0.91**	0.12	0.81
2 (3). When you grow up, will you be a Mommy or a Daddy?	0.04 ± 0.27	**0.76**	0.19	0.72
3 (5). Are there any good things about being a boy?	0.08 ± 0.35	0.43	**0.77**	0.81
4 (6). Is there anything you don't like about being a boy?)[a]	NA	NA	NA	NA
5 (7). Do you think it is better to be a boy or a girl?	0.08 ± 0.35	0.45	**0.82**	0.85
6 (8). In your mind, do you ever think that you would like to be a girl?	0.08 ± 0.36	**0.59**	0.47	0.76
7 (9). In your mind, do you ever get mixed up and you are not really sure if you are a boy or a girl?	0.13 ± 0.44	−0.13	**0.76**	0.35
8 (10). Do you ever feel more like a girl than like a boy?	0.06 ± 0.27	**0.82**	0.20	0.78
9 (11). You know what dreams are, right? When you have a dream at night, are you ever in the dream? If yes, ask: in your dreams, are you a boy, a girl, or sometimes a boy and sometimes a girl?)[a]	NA	NA	NA	NA
10 (12). Do you ever think that you really are a girl?	0.03 ± 0.22	**0.83**	0.22	0.80
11. When you stand in front of the mirror, which part of your body you like the most?[b]	0.07 ± 0.31	0.47	**0.69**	0.79

Bold values indicate the higher factor loadings in one factor compared to the other factor

Factor loadings represent factor loadings after varimax rotation. Number in the brackets refers to numbering applied in the original version of the GIIC

In Zucker et al. (1993), Items 1 and 2 above were loaded onto the cognitive gender confusion factor, whereas Items 4–10 above were loaded onto the affective gender confusion factor. In Wallien et al. (2009) Items 1–10 above loaded into single factor of gender confusion. In our study, Factor 1 = cognitive gender confusion; Factor 2 = affective gender confusion. Cronbach alphas of the Factor 1, Factor 2, and single factor of the Indonesian version of GIIC are: 0.78; 0.86; 0.88, respectively. N = 120

[a] These items were applied in the study but were not included in the principal component analysis and reliability analysis due to large proportion of missing data (*NA* not applicable)

[b] This item was added for this study

Table 3 Mean, SD, and factor loadings of the GIQC items

Original item number	$M \pm SD$ (current study)	Factor loadings reported in studies using GIQC		
		Current study	Johnson et al. (2004)	Elizabeth and Green (1984)
1. Playmate	3.9 ± 1.0	0.62	0.77	0.80
2. Girl's doll	4.3 ± 1.3	0.71	0.74	0.77
3. Boy's doll	4.3 ± 1.2	0.70	0.34	0.30
4. Make-up	4.3 ± 1.3	0.80	0.71	0.77
5. Imitate female model	4.0 ± 1.6	0.75	0.64	0.69
6. Imitate male model	3.0 ± 1.8	−0.47	0.48	0.59
7. Play sports with boys	4.1 ± 1.2	0.69	0.62	0.67
8. Play sports with girls	4.1 ± 1.2	0.63	0.20	0.38
9. Role play[a]	NA	NA	0.89	0.92
10. Girl's type play	4.5 ± 1.0	0.84	0.83	0.88
11. Boy's type play	4.4 ± 0.9	0.80	0.72	0.85
12. Dress-up games*	NA	NA	0.91	0.94
13. Wishes to be the opposite-sex*	4.9 ± 0.5	0.36	0.81	–
14. Stated own self as the opposite-sex*	4.9 ± 0.6	0.23	0.69	–
15. Disliking own sexual parts*	4.7 ± 0.7	−0.09	0.47	–
16. Liking own sexual parts*	1.2 ± 0.7	−0.01	0.02	–

N = 120. Factor loadings represent factor loading after varimax rotation

These items (marked with asterisks) were not applied in this study. Items 9 and 12 had a large missing response; Items 13–16 had poor factor loadings. The remaining 10 items (Items 1–8, 10, and 11) were applied in this study

NA not applicable

Table 4 Median, range, and factor loadings of the gender questionnaire items

Items	Median	Range	Factors	
			1	2
1. During the *past 12 months*, my behavior has been what most people consider appropriate for my sex	1.0	6	0.17	**0.90**
2. During *my lifetime*, my behavior has been what most people consider appropriate for my sex	1.0	6	0.11	**0.91**
3. During *the past 12 months,* I enjoyed being a person of my sex	1.0	6	0.39	**0.66**
4. During *my lifetime,* I enjoyed being a person of my sex	1.0	6	0.39	**0.72**
5. During *the past 12 months,* I have wished I were a person of the opposite sex	1.0	6	**0.76**	0.31
6. During *my lifetime,* I have wished I were a person of the opposite sex	1.0	6	**0.67**	0.47
7. During *the past 12 months,* I have thought I was psychologically a person of the opposite sex	1.0	6	**0.82**	0.37
8. During *my lifetime,* I have thought I was psychologically a person of the opposite sex	1.0	6	**0.76**	0.45
9. During *the past 12 months,* I have been treated as person of the opposite sex by people who knew me well and people who were unfamiliar to me	1.0	6	**0.73**	0.39
10. During *my lifetime,* I have been treated as person of the opposite sex by people who knew me well and people who were unfamiliar to me	1.0	6	**0.68**	0.48
11. During *the past 12 months,* I have presented myself as person of the opposite sex at my job and during leisure time	1.0	6	**0.84**	0.26
12. During *my lifetime,* I have presented myself as person of the opposite sex at my job and during leisure time	1.0	7	**0.82**	0.30
13. During *the past 12 months,* I wanted medical treatment to change my body into a body of the opposite sex	1.0	6	**0.90**	0.08
14. During *my lifetime,* I wanted medical treatment to change my body into a body of the opposite sex	1.0	7	**0.92**	0.09

Bold values indicate the higher factor loadings in one factor compared to the other factor

N = 316. Factor loadings represent factor loading after varimax rotation. Factor 1 = Gender identity and gender role behavior; Factor 2 = Cross-gender identity and cross-gender role behavior. Items 1–8 were obtained from the original version of the Gender Questionnaire (Hines et al., 2003); Items 9–14 were added for this study. The Cronbach's alphas for both factors above were 0.88 and 0.96, respectively

a 9-item single factor PCA model of the GIIC (56.8 % of the total variance explained; total score range: 0 = no gender confusion; 18 = extreme gender confusion).

The Parent-Report Gender Identity Questionnaire for Children (GIQC)

Analysis of the Indonesian version of GIQC showed that the response rates of Items 9 and 12 (original items number) were only 16.7–21.7 %. This suggested that the role-play (Item 9) and the dress-up games (Item 12) were unpopular activities among subjects. In view of the large non-response rates, these two items were excluded and the PCA was performed on 11 items of the Indonesian version of GIQC and generated a one PC solution that explained 37.34 % of the total variance. Table 3 shows the factor loadings of each item obtained in this study in comparison to previous studies on GIQC. The majority of items had similar or higher factor loadings compared to previous studies (Elizabeth & Green, 1984; Johnson et al., 2004), except four items (Items 13–16) that had very low factor loadings. One item (Item 6) had a negative factor loading indicating that reversed scoring was needed. However, this item remained problematic in the reliability analysis; therefore, we excluded this item for further analysis. Internal consistency of the remaining 10 items resulted in a Cronbach's alpha = 0.88 indicating that the Indonesian version of GIQC has a good reliability. The total score of GIQC ranged from 5 to 50. Low scores indicate that parents report more frequent cross-gender behavior in their child (Table 3).

Gender Questionnaire

PCA generated two components explaining 74.9 % of total variance. Table 4 shows that Items 1–4 loaded on Factor 1 and reflected *gender identity* and *gender role behavior*. Items 5–14 loaded on Factor 2 and reflected *cross-gender identity* and *cross-gender role behavior*. The Cronbach's alphas of both factors indicated good internal consistency ($\alpha = 0.88$; $\alpha = 0.96$, respectively). The results were summarized in two sum scores for *gender identity* and *gender role behavior*: in the past 12 months and during the entire lifetime (range sum score: 2–14 for each period) and two sum scores for *cross-gender identity* and *cross-gender role behavior:* in the past 12 months and during the entire lifetime (range sum score: 5–35 for each period). Low scores on gender identity and gender role behavior (Factor 1) indicate a distinct sex-typical gender identity and gender role behavior in the specified periods. Higher scores on cross-gender identity and cross-gender role behavior (Factor 2) indicate a cross-gender identity and more cross-gender role behaviors.

Activities Questionnaire

During the pilot study, two problematic items were identified: Items 13 (degree of girlishness) and Item 14 (degree of boyish

Table 5 Mean, SD, and factor loadings of the Activities Questionnaire items

Items	$M \pm SD$	Factor loadings		
		Feminine	Masculine	Neutral
1. (Male) Playmate	3.1 ± 0.9	−0.47	**0.47**	0.12
2. Dolls	3.0 ± 1.2	**0.79**	−0.20	−0.13
3. Sports	3.6 ± 0.9	0.03	**0.67**	−0.12
4. Cars	3.3 ± 1.1	−0.60	**0.49**	−0.20
5. Play outside	4.1 ± 0.7	−0.05	**0.75**	0.005
6. Drawings	2.7 ± 1.0	0.001	−0.05	**0.71**
7. Rough-tumble play	2.6 ± 1.2	−0.40	**0.65**	−0.11
8. Reading	1.7 ± 0.7	−0.14	0.28	**0.50**
9. Board games	1.9 ± 0.7	−0.34	−0.14	**0.37**
10. Dress-up	2.6 ± 1.2	**0.88**	0.01	−0.02
11. Building blocks	2.4 ± 0.9	0.06	−0.28	**0.66**
12. Cosmetics	2.4 ± 1.2	**0.88**	−0.11	−0.09

Bold values indicate the higher factor loadings in one factor compared to the other factor

N = 254. Factor loadings represent factor loadings after varimax rotation. Items 13 and 14 in the original measure (Hines et al., 2003) were problematic; therefore, both items were not included for PCA and reliability analysis. The Cronbach alphas of the Indonesian version of Activities Questionnaire for the feminine, masculine, and neutral factors were 0.73, 0.87, 0.38 respectively

ness). Due to the absence of appropriate equivalents in local lan guage, we used the terms feminine and masculine. However, all subjects perceived being feminine or masculine referred to having an ideal female or male appearance (as models in commercials). Consequently, these items lack the sensitivity to determine degree of femininity or masculinity and thus we decided to exclude them from further statistical analyses. PCA was performed on the remaining 12 items and generated 3 components of preference explaining 55.2 % of total variance. Table 5 shows that Items 2, 10, and 12 loaded on Factor 1 assessing a feminine type of preferences; Items 1, 3, 4, 5, and 7 loaded on Factor 2 assessing a masculine type of preferences and Items 6, 8, 9, and 11 loaded on Factor 3 assessing a neutral type of preferences. Cronbach's alphas were: feminine $\alpha = 0.87$; masculine $\alpha = 0.73$; and neutral $\alpha = 0.38$. As the neutral scale had poor internal consistency, we report the masculine and feminine scales only. Because feminine preferences were assessed by 3 items and masculine preferences were assessed by 5 items, the score for feminine preferences was multiplied by 1.67 (5/3 = 1.67) to allow a valid comparison. The sum score range was 5–25 for the feminine and masculine components. Higher scores indicate greater preferences for typical feminine or masculine activities.

Method

Study 2: Gender Identity and Gender Role Behavior in Patients with DSD

The study protocol was approved by the board of the ethics committee at the Faculty of Medicine, Diponegoro University, Semarang, Indonesia.

Participants

Patients with DSD

Study participants were recruited from the Sexual Adjustment Team of the Dr. Kariadi Hospital and the Faculty of Medicine, Diponegoro University. This group comprised 118 patients diagnosed with DSD: 60 children (42 boys, 18 girls, ages 6–11 years), 24 adolescents (15 boys, 9 girls, ages 12–17 years), and 34 adults (20 men, 14 women, ages 18–41 years). We excluded individuals with a genital anomaly and dysmorphic features suggestive of malformation syndromes (Hutson, Grover, O'Connell, & Pennell, 2014), patients with sex chromosome DSD without mosaicism, and patients with DSD and intellectual disabilities (indicated from parent reports on their child's academic achievements and/or observed by the physician in interaction with the patient). Of the 168 patients who matched the inclusion criteria, 21 patients (12.5 %) were lost to follow-up due to relocation or invalid contact details and 29 patients (17.3 %) declined to participate. The response rate was 70.2 %. The majority of patients who declined participation were children (51.7 %) who were predominantly male (64.3 %) and who had been diagnosed with 46,XY DSD and hypomasculinization (37.9 %).

Patients with 46,XX DSD were diagnosed with CAH-SV (simple virilizing type of CAH), cloacal exstrophy or gonadal dysgenesis. Patients with 46 XY DSD, suffered from gonadal dysgenesis (diagnosed by hormonal evaluation) or androgen insensitivity syndrome (confirmed by identification of a mutation in the androgen receptor gene). Despite extensive analysis, a molecular diagnosis could not be made in the remaining group of patients with 46 XY DSD and hypomasculinization. Table 7 in Appendix shows the diagnostic characteristics, the reported treatment received, and the gender history of all 118 patients

Table 6 Socioeconomic and cultural background of study participants

Characteristics background	Patients with DSD ($n = 118$)	Matched controls ($n = 118$)	p
Age of study	13.8 ± 7.4	14.2 ± 7.1	n.s.
Region			
Central Java province	100 (84.7)	108 (91.5)	n.s.
Other provinces in Java	12 (10.2)	9 (7.6)	
Outside Java island	6 (5.1)	1 (0.8)	
Ethnicity			
Javanese	108 (91.5)	106 (89.8)	n.s.
Non Javanese	10 (8.5)	12 (10.2)	
Religion			
Islam	112 (94.9)	108 (91.5)	n.s.
Non Islam	6 (5.1)	10 (8.5)	
Education—father[a]	$n = 116$	$n = 114$	
Illiterate	18 (15.5)	15 (13.2)	n.s.
Elementary school	38 (32.8)	31 (27.2)	
High school	49 (42.2)	58 (50.9)	
University education	11 (9.5)	10 (8.8)	
Education—mother[a]	$n = 116$	$n = 117$	
Illiterate	22 (19.0)	14 (12.0)	n.s.
Elementary school	38 (32.8)	34 (29.1)	
High school	48 (41.4)	58 (49.6)	
University education	8 (6.9)	11 (9.4)	
Occupation—father[a]	$n = 116$	$n = 114$	
Unemployed	6 (5.2)	5 (4.4)	.06
Labor	64 (55.2)	46 (40.4)	
Self-employed	19 (16.4)	34 (29.8)	
Staff/Office job	27 (23.3)	29 (25.4)	
Occupation—mother[a]	$n = 116$	$n = 117$	
Unemployed	57 (49.1)	39 (33.3)	.02
Labor	32 (27.6)	35 (29.9)	
Self-employed	12 (10.3)	28 (23.9)	
Staff/Office job	15 (12.9)	15 (12.8)	

Data presented in n (%)

The Fisher's exact test was applied; significant at $p < .05$

Superscript letter 'a' indicates differences in n

who participated in this study. With respect to the measures of gender identity and gender role behavior, assessment was done according to the gender in which the patient was living at the time they participated in the study.

Sixty-one (51.7 %) patients received some surgical or hormonal treatment. In Indonesia, patients are usually informed orally by their physicians. Physicians rarely exchange written medical information and, as a consequence, little is known about past medical examinations, medical diagnostic procedures or medical and surgical treatments. Reports on surgical treatments that were received are shown in Table 7 in Appendix. The remaining 57 patients (48.3 %) had not received any medical or surgical evaluation or treatment prior to study participation. The study included 11 children with a 46,XX karyotype and CAH-SV who had been raised as girls and who had received some hormonal treatment; however, this was often taken irregularly because medication was generally unavailable or unaffordable. There was one boy with a 46, XX karyotype and CAH-SV who had received hormonal treatment for 9 months in infancy, then irregularly for several years, and was left untreated for at least two years prior to this study (S10; Table 7 in Appendix). Due to lack of medical resources, patients with the salt wasting type may not have survived. Two boys diagnosed with gonadal dysgenesis (aged 6; S21; Table 7 in Appendix) and PAIS (aged 9; S32; Table 7 in Appendix) received HCG injections at 1 and 4 years prior to study participation. Twelve girls and 17 boys with various diagnoses (ages 6–11 years)

received genital surgery. The girls were younger at their first surgery than the boys (mean age ± SD for girls: 4.7 ± 2.0 years; for boys 5.2 ± 2.4 years). Twelve boys and 2 girls received multiple genital surgeries.

Matched Controls

This group was composed of 118 healthy adults matched for age (with maximum age disparity of 3 years), gender at the time of study, and residential settings (rural, suburban, or urban). After a potential matched control subject was identified, an invitation to join the study was given. In order to protect patients from being identified by their matched controls, the matched control subjects were informed about the study but only learned that they were selected to participate in a study on gender development carried out by the Faculty of Psychology at the Diponegoro University in Semarang. Control subjects and/or parents of control subjects younger than 18 years who wished to participate gave their written consent prior to assessment.

By design, the patients and the matched control subjects were comparable with respect to socioeconomic and cultural variables. Table 6 summarizes the background of participants in this study. The majority of participants were male, lived in rural areas, came from the Central Java province, were Javanese, and were Moslem. Parents' educational backgrounds varied from illiterate to university level with the majority having attended high school. Most parents worked in the low-income sector or were unemployed, particularly parents of patients with DSD.

Procedure

This psychological study was part of the medical study evaluating the clinical diagnoses of patients with DSD. The psychological follow-up was carried out between March 2007 and May 2011. The DSD diagnosis was based on a physical examination, results of cytogenetic analysis, hormonal data, and molecular analyses. The diagnostic procedures leading to the diagnosis of DSD have been described by Juniarto et al. (2012). Patients were invited to participate in the study and were given oral and written study information by a physician (AZJ). After patients had given written consent, an appointment was made for the gender assessment. The assessment was conducted by a clinical psychologist (AE) in the hospital or at home. She received training to deliver these gender measures and to conduct interviews with patients with DSD. In addition to the measures applied, the history of gender development was also obtained during the interview.

Measures

See Study 1 for a detailed description of the measures applied in this study.

Statistical Analysis

Outcome measures were compared between patients and healthy controls stratified by gender and age. Differences in continuous data with skewed distributions between two groups were summarized as medians (*Mdn*) and tested with the Mann-Whitney *U* test. Differences in categorical data between groups were compared using Fisher's Exact test. Differences between groups were considered significant at $p < .05$ (two-sided). Due to the small number of cases in subgroups, comparisons of different subgroups of DSD diagnoses, or between patients who had changed their gender and patients who did not, were avoided. We display individual data in scatter plots.

Results

Gender Change

Overall, 21 patients changed their gender: 6.7 % (4 of 60) children, 8.3 % (2 of 24) adolescents, and 44.1 % (15 of 34) adults. Gender change was reported by both patients with a 46,XX karyotype (4 patients; 19 %) and a 46,XY karyotype (17 patients; 81 %). In 8 patients (38 %), the parents or physician proposed the gender change, 13 patients (62 %) initiated the gender change (Table 7 in Appendix).

Three patients with a 46,XX karyotype had CAH-SV (S01, S86, S87; siblings). They were assigned female at birth and had changed to be male gradually at ages 2–3. They never had received glucocorticoid treatment. This led to progressive genital and behavioral masculinization and identification as males. The remaining patient with 46,XX karyotype who changed gender was born with cloacal malformation (S92). After birth, she was assigned male without medical evaluation. At age 20, she had entered our hospital. After diagnostic procedures had been carried out, she learned about her karyotype, the development of ovaries, uterus, and female development of body appearance in puberty and decided to continue her life as a woman. She was the only patient in the study who underwent a male-to-female social gender role change.

Among 17 patients with a 46,XY karyotype, parents or health workers had initiated the gender reassignment in four patients (S27, S34, S40, S100) at the ages of 2–8 years. The remaining 13 patients with a 46,XY karyotype took initiatives to change their gender between the ages of 15–27 years. In total, four patients underwent a gender change in about 1 year post study, whereas 17 patients had been living in their changed gender for 2–25 years prior to study participation.

Of these 21 patients who changed gender, 15 patients (10 adults, 5 adolescents) sought medical help at our hospital late in life. Prior to the study, 76.2 % (16 patients) had not received any treatment for their DSD conditions, whereas 23.8% (five patients) had undergone some genital surgeries (Table 7 in Appendix). All 21 patients disclosed their DSD conditions to their parents and/or spouses and

received emotional support and acceptance for their DSD condition and their social gender change. Only two patients had undergone a legal procedure to change their gender on their birth certificates.

Gender Identity Interview for Children (GIIC)

Children with DSD raised as girls reported greater gender confusion than children with DSD raised as boys ($Mdn_{girls} = 0$; $Mdn_{boys} = 0$; $p = .004$). In the matched control groups, the difference in gender confusion between girls and boys was not significant ($Mdn_{girls} = 0$ vs $Mdn_{boys} = 0$). There was a tendency towards significance that children with DSD raised as girls reported greater gender confusion than the matched control girls ($Mdn_{patients} = 0$; vs $Mdn_{controls} = 0$; $p = .08$). Comparison between the DSD and the matched control groups revealed that the gender confusion reported by the 42 children with DSD raised as boys and the 42 matched controls boys ($Mdn_{patients} = 0$ vs $Mdn_{controls} = 0$) did not differ significantly. As shown in Fig. 1, individual differences in GIIC scores between and within groups are present despite the equality of the median values. Of 18 children with DSD raised as girls, 15 had a 46,XX karyotype and CAH-SV. All four children who experienced a gender change (S01, S27, S34, S40) scored higher (meaning more gender confusion) on the GIIC than youngsters who had not experienced a gender change.

Gender Identity Questionnaire for Children (GIQC)

Parents of girls with DSD reported more cross-gender role behavior in their offspring than parents of matched control girls did ($Mdn_{patients} = 21.5$ vs $Mdn_{controls} = 25.5$, $p = .047$). No significant differences were found between parents' reports on boys with DSD and those for control boys ($Mdn_{patients} = 31.0$ vs $Mdn_{controls} = 31.0$). In the DSD group, cross-gender behavior was reported more frequently by parents of girls than parents of boys ($Mdn_{girls} = 21.5$ vs $Mdn_{boys} = 31.0$, $p < .001$). Similarly, among matched controls, parents of girls reported more cross-gender role behavior than parents of boys ($Mdn_{girls} = 25.5$ vs $Mdn_{boys} = 31.0$, $p < .001$).

Figure 2 shows the individual total scores on the GIQC across groups and diagnoses. Parents of girls with 46 XX karyotype and CAH-SV reported less typical gender role behaviors. Eleven of 15 girls with 46,XX karyotype and CAH-SV received genital surgery and/or hydrocortisone medication in the 2–8 years prior to the study. Finally, the girl with a 46,XY karyotype and gonadal dysgenesis who changed her gender to male after she had taken part in this study (S27; Table 7 in Appendix) obtained the lowest scores on the GIQC. Due to small number of cases, we could not perform further comparison analyses to support this finding statistically.

Gender Questionnaire (GQ)

Adolescent girls with DSD reported significantly less sex-typical gender identity and gender role behavior during their lifetime and in the past 12 months than adolescent boys with DSD, during their lifetime and in the past 12 months ($Mdn_{girls} = 2$ vs $Mdn_{boys} = 2$; $p = .02$; $Mdn_{girls} = 2$ vs $Mdn_{boys} = 2$; $p = .01$, respectively). They also reported more cross-gender identity and more cross-gender role behavior during lifetime and in the past 12 months than adolescent boys with DSD ($Mdn_{girls} = 5$ vs $Mdn_{boys} = 5$; $p = .04$; $Mdn_{girls} = 5$ vs $Mdn_{boys} = 5$; $p = .01$, respectively).

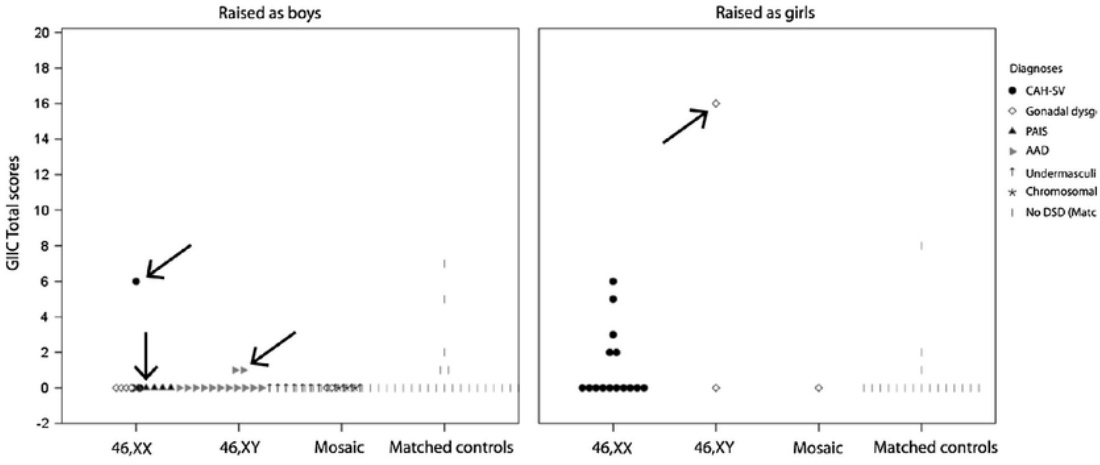

Fig. 1 Gender identity confusion in patients with different karyotypes and DSD diagnoses (GIIC total scores). The plots show individual data (total scores) of the Indonesian version of the GIIC in patients with different karyotypes and DSD diagnoses who were raised as boys or girls in comparison to the matched control boys or girls. Possible score range 0–18; 0 indicates no gender confusion. The higher the scores, the greater gender confusion is. The *arrows* indicate the scores of children who had histories of gender change (3 children—S01; S34; S40—had changed their gender and lived as boys prior to study and one girl—S27—changed her gender to be a boy post study; see Table 7 in Appendix for details)

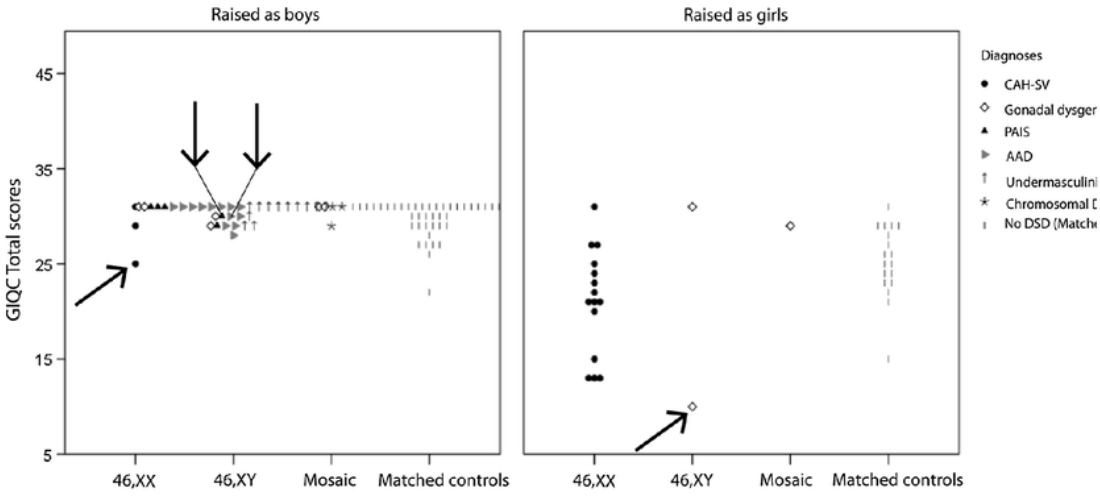

Fig. 2 Sex-typical gender role behavior in patients with different karyotypes and DSD diagnoses (GIQC total scores). The plots display the individual data of the GIQC in patients with different karyotypes and DSD diagnoses, raised as boys or girls, in comparison to the matched control children (no DSD). Possible total score range 5–50. Lower scores indicate less sex-typical gender role behavior in children. The *arrows* indicate the scores of children who had histories of gender change (3 children—S01; S34; S40—had changed their gender and lived as boys prior to study and one girl—S27—changed her gender to be a boy post study; see Table 7 in Appendix for details)

Adult men with DSD scored higher than control men on the lifetime scales, but not on the past-12 months scales, of the GQ, meaning that they were less sex-typical in identity and behavior and had more problems related to their gender identity and behavior (gender identity and gender role behavior: $Mdn_{\text{patients}} = 2.5$ vs $Mdn_{\text{controls}} = 2$; $p = .01$; cross-gender identity and behavior in the past: $Mdn_{\text{patients}} = 6.5$ vs $Mdn_{\text{controls}} = 5$; $p = .01$). The majority of men with DSD (13 of 20 or 65%; 2 had a 46,XX CAH-SV and 11 had a 46,XY karyotype; see Table 7 in Appendix) had initially been raised as girls but had changed their gender to male later in life. In contrast, adult women with DSD did not differ significantly from the matched control women in their gender identity and gender role behavior during their lifetime and in the past 12 months ($Mdn_{\text{patients}} = 2$ vs $Mdn_{\text{controls}} = 2$; $p = .77$; $Mdn_{\text{patients}} = 2$ vs $Mdn_{\text{controls}} = 2$), nor were differences evident in their cross-gender identity and cross-gender role behavior during their lifetime and in the past 12 months ($Mdn_{\text{patients}} = 5$ vs $Mdn_{\text{controls}} = 5$; $Mdn_{\text{patients}} = 5$; $Mdn_{\text{controls}} = 5$). Among the adult women with DSD, 3 had a 46,XX karyotype, 3 had mosaic sex chromosome, and 8 had a 46,XY karyotype, but 2 of them changed their gender later (see Table 7 in Appendix).

Of 24 adolescents and 34 adults with DSD who completed the GQ, there were 2 adolescents and 15 adults who had a history of gender change. The individual total scores on the GQ scales among patients with different karyotypes are shown in Fig. 3a–d. Of these 17 patients who had a history of gender change, 3 (S80, S97, and S98; Table 7 in Appendix) were living as females when they participated in the study but then changed their gender in the following years. On the GQ for females, they obtained high score on all scales (see patients indicated by arrows in Fig. 3a–d). This is in contrast to the majority of patients who had changed their gender prior to study participation. These men scored high on the lifetime-scales, but scored low on the past 12 months-scales, indicating they experienced less gender dysphoria after their gender change than before.

We compared patients who had received surgical and hormonal treatment with patients who had not. Due to the small sample of treated and untreated groups, we combined GQ data from adolescents and adult patients to enhance the comparison analysis and found significant differences in females, but not in males. On all scales, adolescents and adults females with DSD who had never received any treatment indicated significantly more gender dysphoria than women who had received some treatments in their measures of gender identity and gender role behavior during their lifetime and in the past 12 months ($Mdn_{\text{untreated}} = 4.0$ vs $Mdn_{\text{treated}} = 2.0$; $p = .01$; $Mdn_{\text{untreated}} = 4.0$ vs $Mdn_{\text{treated}} = 2.0$; $p = .02$, respectively) and in cross-gender identity and cross-gender role behavior during their lifetime and in the past 12 months ($Mdn_{\text{untreated}} = 7.0$ vs $Mdn_{\text{treated}} = 5.0$; $p = .01$; $Mdn_{\text{untreated}} = 6.0$; $Mdn_{\text{treated}} = 5.0$; $p = .02$, respectively). Figure 4 shows individual data on GQ between treated and untreated males and females with DSD, adolescents and adults together, in view of their gender change history. Among the females who indicated gender dysphoria, 3 changed gender post-study (indicated by arrows in Fig. 4a–d; see S80, S97, and S98 in Table 7 in Appendix). The patient with a 46,XX karyotype and cloacal malformation who underwent a male-to-female gender change after participation reported less severe gender dysphoria than the 3 girls who underwent a female-to-male gender change after participation. Among the males who changed their gender from female to male prior to enrolling in the study, 5

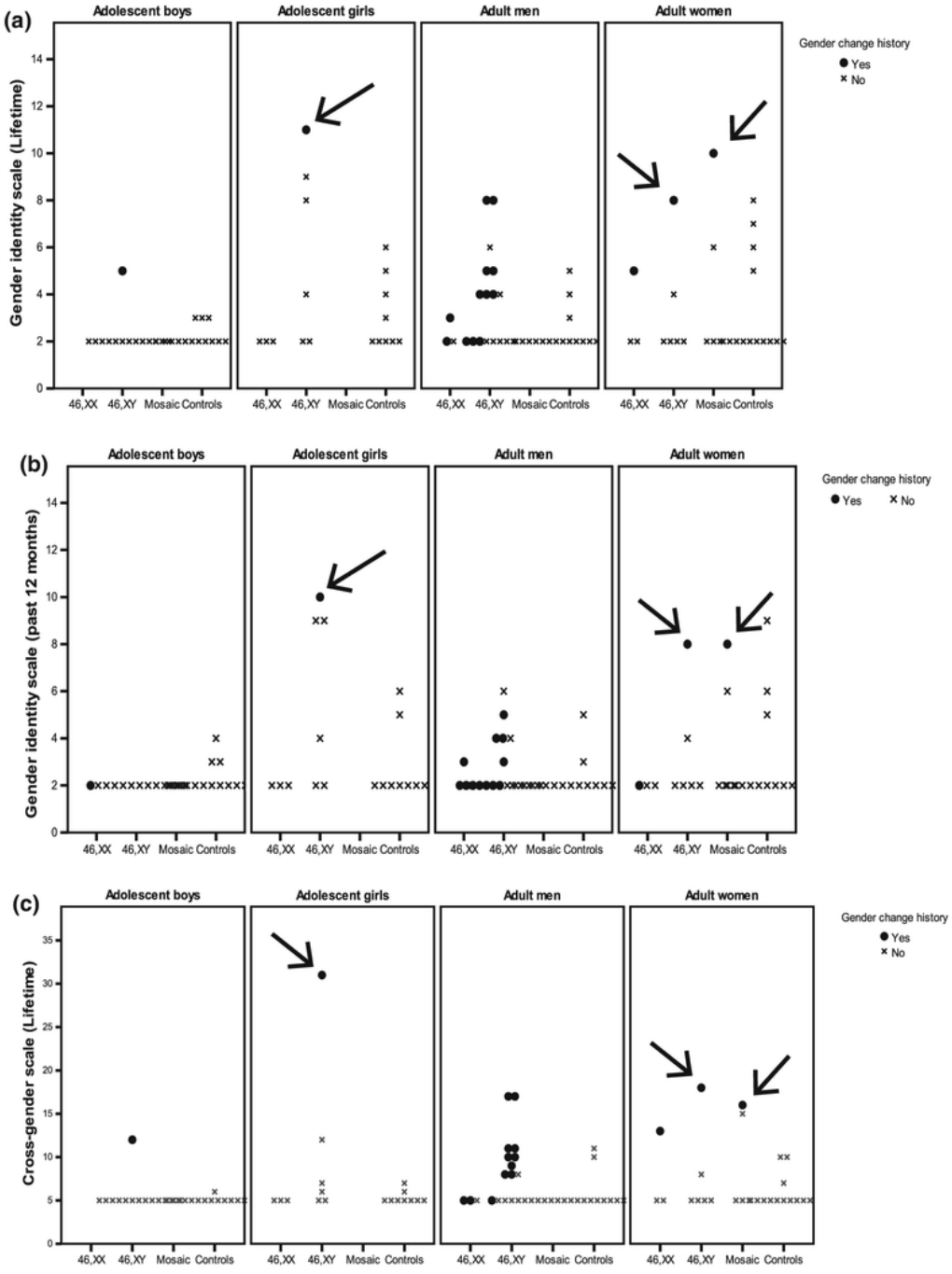

Fig. 3 Gender identity and gender role behavior in patients with different karyotype and gender change history (GQ data). These plots **a**, **b** display individual scale score on the gender identity and behavior reported during lifetime (**a**) and in the past 12 months (**b**). Possible scale score range 2–14. Higher scores indicate less sex-typical gender identity and behavior reported during lifetime or in the past 12 months. The arrows indicate patients who changed gender post study (S80, S97, S98; Table 7 in Appendix). These plots (**c**, **d**) display individual scale score on the cross-gender identity and behavior reported during lifetime (**c**) and in the past 12 months (**d**). Possible scale score range 5–35. Higher scores indicate more cross-gender identity and behavior reported during lifetime or in the past 12 months. The *arrows* indicate patients who changed gender post study (S80, S97, S98; Table 7 in Appendix)

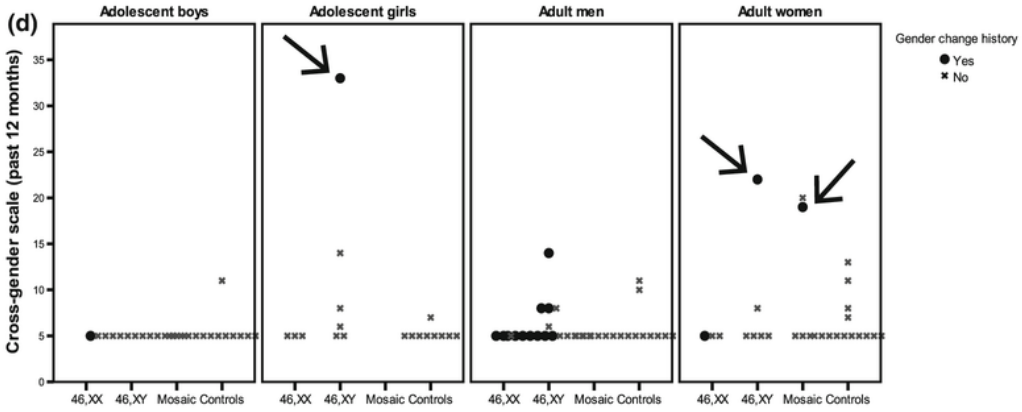

Fig. 3 continued

received some treatment and 8 never received any medical treatment prior to this study.

Activities Questionnaire (AQ)

Figure 5 shows the findings on the Activities Questionnaire. Adolescent boys and girls, in either the DSD or the matched control group, recalled different preferences in childhood activity. Among adolescents with DSD, boys recalled more masculine activities than girls did ($Mdn_{boys} = 20$ vs $Mdn_{girls} = 16.5$; $p < .001$), whereas girls recalled more feminine activities than boys did ($Mdn_{girls} = 10$ vs $Mdn_{boys} = 5$; $p = .002$). Similar findings were reported among the matched control adolescents: ($Mdn_{boys} = 19$ vs $Mdn_{girls} = 14.5$; $p = .008$), ($Mdn_{girls} = 14.2$ vs $Mdn_{boys} = 5$; $p = .001$).

Adolescent girls with DSD did not differ from the matched control girls in recalled masculine and feminine activities in childhood ($Mdn_{patients} = 16.5$ vs $Mdn_{controls} = 14.5$ and $Mdn_{patients} = 10$ vs $Mdn_{controls} = 14.2$). Similarly, adolescent boys with DSD did not differ from the matched control boys in recalled masculine and feminine activities in childhood ($Mdn_{patients} = 20$ vs $Mdn_{controls} = 19$; $p = .42$ and $Mdn_{patients} = 5$ vs $Mdn_{controls} = 5$).

Men and women in both the DSD and matched control groups, recalled differences in the childhood activities they preferred. Among adults with DSD and matched controls, men recalled more masculine activities than women ($Mdn_{men} = 18$ vs $Mdn_{women} = 14.5$; $p < .001$ and $Mdn_{men} = 19$ vs $Mdn_{women} = 13$; $p < .001$, respectively), whereas women recalled more feminine activities than men ($Mdn_{women} = 11.7$ vs $Mdn_{men} = 5.0$; $p < .001$ and $Mdn_{women} = 13.4$ vs $Mdn_{men} = 5.0$; $p < .001$, respectively).

Women with DSD did not differ from the matched control women in recalled masculine and feminine type of childhood activities ($Mdn_{patients} = 14.5$ vs $Mdn_{controls} = 13$; $p = .45$ and $Mdn_{patients} = 11.7$ vs $Mdn_{controls} = 13.4$). Men with DSD do not differ from the matched control men in recalled masculine and feminine type of childhood activities ($Mdn_{patients} = 18$ vs $Mdn_{controls} = 1!9$; $p = .76$ and $Mdn_{patients} = 5$ vs $Mdn_{controls} = 5$).

Discussion

This study aimed to investigate the gender development of Indonesian patients with DSD who, for some years, had not received a diagnostic workup or treatment, or received limited treatment. To our knowledge, this is the first study reporting gender development in patients with DSD in Indonesia and also the first study to consider gender development in a large sample of patients with DSD who did not receive medical or surgical treatment.

Gender Change

Most studies of gender change during puberty report on patients diagnosed with 5α-reductase 2 and 17β-hydroxy-steroid dehydrogenase 3 deficiencies. Gender reassignment and self-initiated gender change in children and adolescents with DSD has been reported in several countries: Dominican Republic, (Imperato McGinley et al., 1979), Mexico, (Méndez et al., 1995), Brazil (Mendonca et al.,1987, 2000, 2003), India (Ammini et al., 2002; Gupta et al., 2010), the United States (Reiner, 2005), China (Jingde et al., 2009), and Egypt (Ismail & Mazen, 2010). However, these studies were conducted on patients who received medical and surgical treatment.

We observed that among late diagnosed and untreated patients with DSD, there was a substantial number ($n = 21$) who had undergone a gender reassignment or changed their gender. The majority of these patients (81 %) had 46,XY DSD and hypomasculinization due to different causes. Only one patient, with 46,XX cloacal exstrophy, changed her gender from male to female. Most patients developed a wish to change their gender during puberty or adulthood (aged 15 or older). In three siblings (Table 7 in Appendix: patients S01, S86 and S87), a gender reassignment

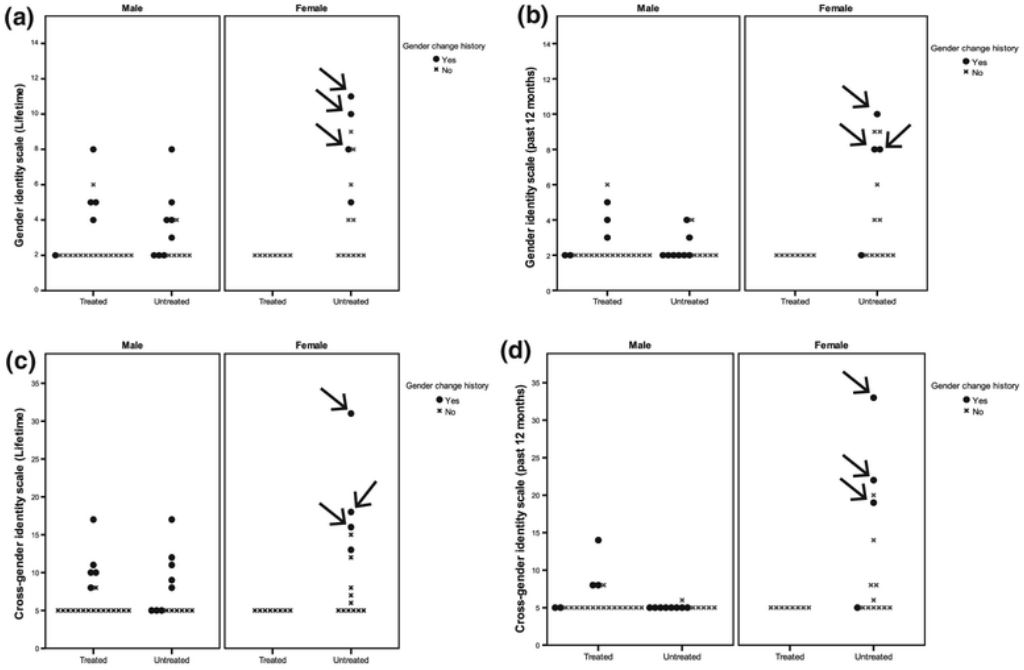

Fig. 4 Comparison on GQ data between treated and untreated adolescents and adult patients. The plots display individual total scores of patients in the gender identity and gender role behavior reported during lifetime (**a**) and in the past 12 months (**b**). Possible score range 2–14. Higher scores indicate less sex-typical gender identity and behavior reported during lifetime or in the past 12 months. The *arrows* indicate patients who changed their gender post study (S80, S97, S98; Table 7 in Appendix). The plots **c**, **d** display individual total scores of patients in the cross-gender identity and cross-gender role behavior reported during lifetime (**c**) and in the past 12 months (**d**). Possible score range 5–35. Higher scores indicate more cross-gender identity and cross-gender role behavior reported during lifetime or in the past 12 months. Comparison analysis revealed significant differences in females, but not in males, in all scales (**a**–**d**). Compared to girls with DSD who had received some treatment, untreated girls with DSD reported less feminine gender-identity and gender role behavior and more often reported cross-gender identity and cross-gender role behavior during lifetime and in the past 12 months

happened gradually at an earlier stage in life (aged 2–3). These patients had 46,XX DSD due to CAH-SV, were assigned female at birth, but had never received glucocorticoid treatment.

The remaining 21 patients with a 46,XX karyotype and CAH-SV did not report problems in gender identity. Two were assigned male at birth, never received glucocorticoid, and were still living in the male gender. Nineteen were assigned female at birth, received glucocorticoid treatment and genital surgery, and were identified as female. In many patients, gender was assigned by parents, midwives, or local medical health workers without the help of any diagnostic evaluation. We do not know which criteria they used in the decision to raise the neonate as a boy or a girl. By absence of medical diagnoses, we assume that gender assignment had been done based on genital phenotype and/or parental wishes. We assume that in patients with 46,XY DSD and hypomasculinization at birth, the masculinization process at puberty influenced the later development of a male gender and, consequently, a wish to change gender from female to male (Sisk & Zehr, 2005). The development of a male identity may result from direct effects of androgens on brain areas important to gender development. Alternatively, perceived changes in the body, which are contrary to the expected pubertal body changes, may introduce gender confusion. On many occasions, patients were misidentified as transsexuals and named *Waria* or *Banci* (local terms for male-to-female person with transgender). Treatments to reverse established changes of masculinization are often too expensive for poor patients to consider. For these individuals, there may be social and economic advantages to live as men.

This study demonstrates that many, but not all, adults with DSD in Indonesia experienced long-term gender problems and this was particularly evident in those patients whose behavior and interests were not in line with their assigned gender (see findings from the Gender Questionnaire) and whose bodies developed atypical secondary sex characteristics at puberty (see Table 7 in Appendix for patients with history of gender change). It is striking that changes in gender are particularly obvious among

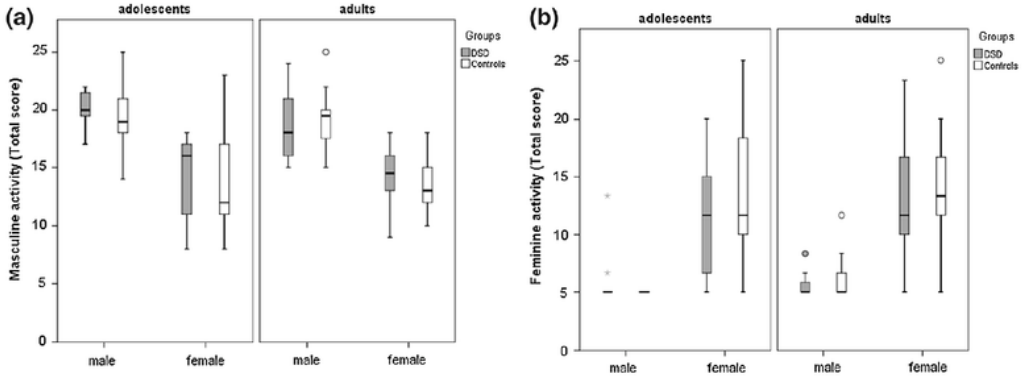

Fig. 5 Recalled childhood activities (the AQ data) reported by adolescents and adults on **a** masculine and **b** feminine type of childhood activities. Patients with DSD did not differ from the matched controls in recalled childhood activity. Either in adolescent or adult group, males recalled more masculine type of childhood activity than females did; females recalled more feminine activity than males did. Females preferred both masculine and feminine type of activities; whereas males preferred masculine activities only

patients living in poverty or in regions in which medical knowledge and help are limited. From Table 7 in Appendix, it can be inferred that in a substantial number of patients, gender-related problems were evident before patients took the decision to change gender.

Gender Identity and Gender Role Behavior in Children

On the measures for gender identity and gender role behavior, children with DSD raised as girls experienced greater gender confusion than the matched control girls or children with DSD raised as boys. The parents of girls with DSD reported more masculine behavior in their daughters than parents of the matched control girls did. The majority of these girls had 46,XX karyotype and CAH. Our findings are in line with previous studies conducted in Western countries that reported tomboyish behavior in girls with CAH (Berenbaum, 1999; Berenbaum et al., 2000; Collaer & Hines, 1995; Meyer-Bahlburg et al., 2004; Pasterski et al., 2014; Zucker et al., 1993). Girls showed a wider range of preferred activities and play behavior than boys whereas boys seemed to narrow their focus on sex-typical activities and gender role behavior.

Gender Identity and Gender Role Behavior in Adolescents

Adolescent girls with DSD reported more dissatisfaction with their assigned gender and had higher scores on cross-gender identity/behavior than adolescent boys with DSD. These differences were not observed among matched control boys and girls. Moreover, adolescents with DSD and the matched controls reported similarities in recalled childhood activities. Boys predominantly preferred masculine activities only whereas girls recalled both masculine and feminine activities.

Gender Identity and Gender Role Behavior in Adults

Adults with DSD living as men at the time of study reported more dissatisfaction with their assigned gender, more often identified oneself in the opposite gender, and more often displayed cross-gender behavior during lifetime than the matched control men. These differences, however, were not reported in relation to the period of the past 12 months. It is important to remember that the majority of men with DSD in this study had undergone a female-to-male gender change and had been living as male for 2–25 years before this study. Thus, they reported their dissatisfaction, experiences, and gender-related problems while living as women.

The Activities Questionnaire could distinguish well between typically male and female childhood activities. Comparison between patients and matched controls revealed no significant differences in recalled feminine or masculine type of childhood activities. Our findings were similar to a previous study conducted in a Western country (Hines et al., 2003), although our study comprised many patients with a history of gender change whereas the Hines et al. study did not.

Treatment versus No Treatment

We investigated differences in gender-related problems between treated and untreated patients and observed differences in females, but not in males. Adolescent girls and women with DSD who never received any medical treatment for their DSD condition reported

more dissatisfaction with their gender identities and greater cross-gender role behavior than adolescent girls and women with DSD who had received some treatment.

Study Limitations

With respect to the utilization of Western measures, we learned that several questions from the Western measures need to be applied carefully and considered for application in non-Western cultures. One has to be cautious when the comparing the psychometric properties of the Indonesian instruments with the European and North American instruments since differences in the composition of the study samples may affect the psychometric results obtained. Future improvement on these measures should include local concepts of femininity and masculinity, gender role behavior, and the social-cultural context of the study sample. Additionally, the lack of uniformity in clinical management in participants likely contributed to variation in results.

Conclusion

Similar to studies performed in Western societies, masculine gender role, but not identity, is the norm for girls with CAH in Indonesia. This study also revealed gender-related problems among late diagnosed patients with DSD, particularly among patients who have been raised as females. It also revealed a large number of patients who underwent female-to-male gender reassignment or self-initiated gender change. Such change most often occurred in females with 46,XY DSD and with 46,XX CAH who did not receive medical care. This argues for an important role for postnatal androgen action in human gender development.

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Appendix

See Table 7.

Table 7 Characteristics of patients with DSD: diagnoses, reported treatment received, and gender development history ($n = 118$)

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study	Gender development history				
				Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]	
Children (S01–60)														
S01	6	46,XX DSD	CAH-SV[a]	3	2.8	6	3	F	None	F	F→M (age 2–3)	M	Gradually. Self-initiated gender change. Sibling of S86 and S87	
S02	6	46,XX DSD	CAH-SV[a]	3	4.5	4	4	F	Hydrocortisone since age 3	F	F	F	No	
S03	6	46,XX DSD	CAH-SV[a]	6	4	4	4	F	Clitoridoplasty age 6, Hydrocortisone since age 6	F	F	F	No	
S04	6	46,XX DSD	CAH-SV[a]	4	4	7	4	F	Hydrocortisone since infancy; clitoridoplasty age 5	F	F	F	No	
S05	6	46,XX DSD	CAH-SV[a]	6	4.5	4	4	F	Hydrocortisone since age 4 (other clinic)	F	F	F	No	
S06	7	46,XX DSD	CAH-SV[a]	2 months	2.5	4	4	F	Hydrocortisone since age 3	F	F	F	No	
S07	8	46,XX DSD	CAH-SV[a]	4	4.1	4	4	F	Medication at birth (other clinic; details unknown); clitoridectomy age 5	F	F	F	No	

Table 7 continued

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study	Gender development history			
				Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]
S08	8	46,XX DSD	CAH-SV[a]	2	3	4	3	F	Hydrocortisone since age 1; clitoridectomy age 2	F	F	F	No
S09	8	46,XX DSD	CAH-SV[a]	8	4.3	4	4	F	None	F	F	F	No
S10	8	46,XX DSD	CAH-SV[a]	8	6	4	5	M	Hydrocortisone irregularly since infancy	M	M	M	No
S11	8	46,XX DSD	CAH-SV[a]	8	5.5	3	3	M	None	M	M	M	No
S12	9	46,XX DSD	CAH-SV[a]	7	4.5	4	4	F	None	F	F	F	No
S13	10	46,XX DSD	CAH-SV[a]	6	4.5	4	3	F	Hydrocostisone irregularly at infancy; regularly since age 4; Clitoridoplasty age 7	F	F	F	No
S14	11	46,XX DSD	CAH-SV[a]	7	5	4	4	F	None	F	F	F	No
S15	11	46,XX DSD	CAH-SV[a]	11	3	7	4	F	Clitoridectomy age 2; hydrocortisone since age 11	F	F	F	No
S16	11	46,XX DSD	CAH-SV[a]	11	Normal clitoris	1	3	F	None	F	F	F	No
S17	11	46,XX DSD	CAH-SV[a]	11	Normal clitoris	4	2	F	Clitoridectomy age 4 (other clinic)	F	F	F	No
S18	11	46,XX DSD	CAH-SV[a]	11	3.5	4	3	F	Clitoridoplasty age 8 (other clinic) and 11; hydrocortisone age 11	F	F	F	No
S19	7	Chromosomal DSD	46,XX/47,XY	1	3	9	4	M	None	M	M	M	No
S20	7	Chromosomal DSD	46,XX/47,XY	7	3	8.5	4	M	None	M	M	M	No
S21	9	Chromosomal DSD	46,XY/46,XX	4	3.5	6	2	M	Orchidectomy age 4; HCG injection age 5	M	M	M	No
S22	6	Chromosomal DSD	46,XY,r(Y)/45,X[c]	2	1.5	6	2	M	Surgery once (details unknown)	M	M	M	No
S23	7	Chromosomal DSD	46,XY/45,X	2	1.7	1	4	F	Vaginoplasty, orchidectomy age 4	F	F	F	No
S24	9	Chromosomal DSD	46,XY/45,X	9	4	5	3	M	None	M	M	M	No
S25	6	46,XY DSD	GD[b]	2	2	5	2	M	None	M	M	M	No
S26	6	46,XY DSD	GD[b]	5	3	5	3	M	Chordaectomy age 5; Urethroplasty age 6	M	M	M	No
S27	7	46,XY DSD	GD[b]	3	3	9	3	M	Surgery age 2 (details unknown; other clinic)	M	M	M	No
S28	8	46,XY DSD	GD[b]	7	3.5	3	3	F	None	F	F→A	A	Male gender reassignment post study
S29	10	46,XY DSD	GD[b]	10	Normal clitoris	1	6	F	None	F	F	F	No
S30	11	46,XY DSD	GD[b]	10	4.5	6	2-3	M	None	M	M	M	No
S31	6	46,XY DSD	PAIS[c]	2	2	6	2	M	Surgery (details unknown)	M	M	M	No
S32	6	46,XY DSD	PAIS[c]	5	2.5	7	3	M	HCG injection age 5	M	M	M	No

Table 7 continued

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study	Gender development history			
				Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]
S33	10	46,XY DSD	PAIS[e]	4	4.9	10	2	M	None	M	M	M	No
S34	10	46,XY DSD	PAIS[e]	10	4.5	6.5	4	M	None	F	F→M (age 3)	M	Gender reassignment (parents)
S35	11	46,XY DSD	PAIS[e]	11	4.5	6	3	M	None	M	M	M	No
S36	6	46,XY DSD	Hypomasculinization[d]	1	2	6	3	M	Hypospadia correction twice, urethroplasty age 2	M	M	M	No
S37	6	46,XY DSD	Hypomasculinization[d]	3	2	6	3	M	Hypospadia corrections three times (details unknown)	M	M	M	No
S38	6	46,XY DSD	Hypomasculinization[d]	4	3	5	2	M	Chordaectomy, orchidopexy age 4; urethroplasty age 5	M	M	M	No
S39	6	46,XY DSD	Hypomasculinization[d]	6	3	6	3	M	None	M	M	M	No
S40	7	46,XY DSD	Hypomasculinization[d]	2	3.7	5.5	4	F	None	F	F→M (age 2)	M	Gender reassignment
S41	8	46,XY DSD	Hypomasculinization[d]	7	3	4	2	M	None	M	M	M	No
S42	8	46,XY DSD	Hypomasculinization[d]	3	3	6	2	M	Chordaectomy age 2; urethroplasty age 3 and 4 (other clinic)	M	M	M	No
S43	9	46,XY DSD	Hypomasculinization[d]	9	3	6	3	M	None	M	M	M	No
S44	10	46,XY DSD	Hypomasculinization[d]	7	2	6	3	M	Urethroplasty, chordaectomy, age 7 and 10	M	M	M	No
S45	10	46,XY DSD	Hypomasculinization[d]	8	2.5	6	3	M	Surgery age 9 and 10 (details unknown)	M	M	M	No
S46	11	46,XY DSD	Hypomasculinization[d]	7	3	6	3	M	Orchidopexy age 7	M	M	M	No
S47	11	46,XY DSD	Hypomasculinization[d]	11	3.5	6	4	M	None	M	M	M	No
S48	11	46,XY DSD	Hypomasculinization[d]	11	4	6	3	M	None	M	M	M	No
S49	6	46,XY DSD	Hypomasculinization[d]	2	4	9	3	M	Surgery twice (details unknown; other clinic)	M	M	M	No
S50	6	46,XY DSD	Hypomasculinization[d]	5	3	9	3	M	Chordaectomy, urethroplasty age 5	M	M	M	No
S51	6	46,XY DSD	Hypomasculinization[d]	6	2.4	9	2	M	None	M	M	M	No
S52	7	46,XY DSD	Hypomasculinization[d]	7	3	9	2	M	None	M	M	M	No
S53	8	46,XY DSD	Hypomasculinization[d]	3	3	10	2	M	None	M	M	M	No
S54	9	46,XY DSD	Hypomasculinization[d]	4	2.5	9	3	M	Hypospadia correction age 6	M	M	M	No
S55	9	46,XY DSD	Hypomasculinization[d]	6	3	9	2	M	None	M	M	M	No
S56	9	46,XY DSD	Hypomasculinization[d]	6	3 (low position)	10	3	M	Surgery age 6, 7, 8 (details unknown)	M	M	M	No
S57	9	46,XY DSD	Hypomasculinization[d]	9	2.7	10	3	M	None	M	M	M	No
S58	9	46,XY DSD	Hypomasculinization[d]	8	3.5	9	3	M	None	M	M	M	No

Table 7 continued

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study	Gender development history			
				Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]
S59	10	46,XY DSD	Hypomasculinization[d]	7	4	9	3	M	Hypospadia correction age 4, 6, 7	M	M	M	No
S60	10	46,XY DSD	Hypomasculinization[d]	9	3.5	9	3	M	Chordaectomy age 9	M	M	M	No
Adolescents (S61–84)													
S61	12	46,XX DSD	CAH-SV[a]	12	normal clitoris	1	3	F	Clitoridectomy age 3 (other clinic)	F	F	F	No
S62	16	46,XX DSD	CAH-SV[a]	16	normal clitoris	1	2	F	Clitoridectomy age 7 (other clinic).	F	F	F	No
S63	13	46,XX DSD	Cloacal mal-formation[e]	12	1.9	1	0	F	Colostomy repair one day after birth	F	F	F	No
S64	13	Chromosomal DSD	46,XY/45,X	12	3	8	2	M	Hypospadia correction, gonadectomy age 12	M	M	M	No
S65	14	Chromosomal DSD	46,XY/45,X	10	3.2	10	1	M	None	M	M	M	No
S66	14	Chromosomal DSD	46,XY/45X	13	4	8	2	M	Chordectomy age 13	M	M	M	No
S67	13	46,XY DSD	GD[b]	12	4.1	8	3	F	None	F	F	F	No
S68	15	46,XY	GD[b]	15	6.5	8	2	M	None	F	F→M	M	Doubt own gender at age 12. Self-initiated gender change
S69	17	46,XY DSD	GD[b]	13	Normal clitoris	1	6	F	Gonadectomy age 10 (other clinic); Estrogen supplement since age 13	F	F	F	No
S70	17	46,XY DSD	GD[b]	17	5	4	4	F	None	F	F	F	No
S71	16	46,XY DSD	CAIS[c]	16	Normal clitoris	1	6	F	None	F	F	F	No
S72	12	46,XY DSD	PAIS[c]	5	1.5	7	4	M	None	M	M	M	No
S73	12	46,XY DSD	PAIS[c]	9	3	5	3	F	None	F	F	F	No
S74	13	46,XY DSD	PAIS[c]	12	3.1	6	3	M	None	M	M	M	No
S75	15	46,XY DSD	PAIS[c]	12	3.1	6	5	M	Testosterone injection, gynecomastia age 13; hydrocele surgery age 14	M	M	M	No
S76	12	46,XY DSD	Hypomasculinization[d]	12	5	6	3	M	None	M	M	M	No
S77	13	46,XY DSD	Hypomasculinization[d]	13	5	6	3	M	Chordectomy age 5, 6	M	M	M	No
S78	14	46,XY DSD	Hypomasculinization[d]	14	3.2	7	3	M	Surgery age 4 (details unknown); chordectomy, urethroplasty age 14	M	M	M	No
S79	14	46,XY DSD	Hypomasculinization[d]	10	3	5	2	M	Urethroplasty age 8	M	M	M	No
S80	17	46,XY DSD	Hypomasculinization[d]	12	7.1	6,5	3	F	None	F	F→A (age 18)	A	Male gender reassignment post study
S81	17	46,XY DSD	Hypomasculinization[d]	14	6.5	6	2	M	Hypospadia correction age 15	M	M	M	No
S82	13	46,XY DSD	Hypomasculinization[d]	8	4	9	2	M	Surgery (details unknown)	M	M	M	No

Table 7 continued

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study	Gender development history			
				Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]
S83	14	46,XY DSD	Hypomasculinization[d]	9	5.2	9	2	M	Chordectomy, hypospadia correction age 10 and 11	M	M	M	No
S84	15	46,XY DSD	Hypomasculinization[d]	14	5	9	2	M	Chordectomy age 14, urethroplasty age 15	M	M	M	No
Adults (S85–118)													
S85	18	46,XX DSD	CAH-SV[a]	11	3.6	4	3	F	Clitoridectomy age 16; Hydrocortisone	F	F	F	No
S86	22	46,XX DSD	CAH-SV[a]	17	7	4	5	M	None	F	F→M (age 2–3)	M	Gradually. Self-initiated. CAH identified in adulthood. Sibling of S01 and S87
S87	24	46,XX DSD	CAH-SV[a]	24	5	7	5	M	None	F	F→M (age 2–3)	M	Sibling of S01 and S86. See remarks S86
S88	36	46,XX DSD	CAH-SV[a]	33	3.6	4	3	F	Clitoral reduction age 34; Prednisone since age 36	F	F	F	No
S89	22	46,XX DSD	Cloacal mal-formation[e]	15	3.5	4	2	M	None	M	M→F (age 20)	F	Self-initiated gender change
S90	18	46,XX DSD	GD[b]	10	3.2	6	4	M	Treatment in other clinic (unknown), in infancy and age 7	M	M	M	No
S91	18	Chromosomal DSD	46,Xi(X)(q10)/45,X	18	Normal clitoris	1	0	F	None	F	F	F	No
S92	20	Chromosomal DSD	46,Xi(X)(q10)(85 %)/45,X(15 %)	20	Normal clitoris	1	0	F	Pills (unknown; to get menstruation) age 20; Dianne35 pills	F	F	F	No
S93	29	Chromosomal DSD	45,X(99 %), 46,XX,iXq(1 %)	25	Normal clitoris	1	0	F	Cycloprogynova age 23; Profertil, Zumenon age 30	F	F	F	No
S94	19	Chromosomal DSD	46,XY/45,X	19	5.5	6	3	F	None	F	F	F	No
S95	20	Chromosomal DSD	46,XidicY	20	6	4	3	F	None	F	F→A (age 13)	A	Started living as male about one year after the study (age 21)
S96	19	46,XY DSD	GD[b]	14	4.1	4	3	F	Gonadectomy age 14	F	F	M	Self-initiated gender change. Just started living as male
S97	19	46,XY DSD	GD[b]	19	Normal clitoris	2	6	F	None	F	F	F	No
S98	19	46,XY DSD	GD[b]	19	5	8	3	F	None	F	F→A	A	Started living as male about one year after the study
S99	21	46,XY DSD	GD[b]	14	6	5	2	F	None	F	F→M (age 14)	M	Self-initiated gender change

Table 7 continued

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study	Gender development history			
				Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]
S100	21	46,XY DSD	GD[b]	21	5	4	3	F	Chordectomy age 11	F	F→M (age 7)	M	Gender reassignment (parents)
S101	23	46,XY DSD	GD[b]	23	Normal clitoris	1	7	F	None	F	F	F	No
S102	27	46,XY DSD	GD[b]	27	5	4.5	3	F	None	F	F	F	No
S103	27	46,XY DSD	GD[b]	26	2.5	3	4	M	None	M	M	M	No
S104	39	46,XY DSD	GD[b]	39	1	5	5	F	None	F	F	F	No
S105	41	46,XY DSD	GD[b]	41	3	6	4	M	None	F	F→M (age 16)	M	Doubt own gender identity aged 8. Self initiated gender change. Married
S106	24	46,XY DSD	CAIS[c]	24	Normal clitoris	1	6	F	None	F	F	F	No
S107	18	46,XY DSD	PAIS[e]	11	3	5	3	M	None	M	M	M	No
S108	18	46,XY DSD	PAIS[e]	15	6.3	7	3	M	Chordectomy age 15; urethroplasty age 16	M	M	M	No
S109	20	46,XY DSD	PAIS[e]	16	3.7	9	2	M	Gynecomasty, chordectomy, hypospadia corrections age 14 and 15	M	M	M	No
S110	27	46,XY DSD	PAIS[e]	27	4.5	6	4	F	None	F	F→M (age 24)	M	Self-initiated gender change. Relocation to other island and start living as male
S111	31	46,XY DSD	PAIS[e]	26	2.6	6	3	F	Gynecomasty, chordee correction age 24; Hypospadia correction age 26	F	F→M (age 27)	M	Gender ambiguity at admission. Change gender with medical consultancy. Married; adopted a child
S112	19	46,XY DSD	Hypomasculinization[d]	14	5.3	7	2	M	Chordectomy age 15; urethroplasty age 19	M	M	M	No
S113	22	46,XY DSD	Hypomasculinization[d]	17	1.5	3	3	F	None	F	F→M (age 19)	M	Self-initiated gender change
S114	26	46,XY DSD	Hypomasculinization[d]	23	5.9	5	3	F	Gynecomasty, chordectomy, hypospadia correction age 23	F	F→M (age 24)	M	Gender ambiguity at admission. Change gender after medical consultancy. Married
S115	20	46,XY DSD	Hypomasculinization[d]	15	6	11	2	M	Hypospadia correction age 5	M	M	M	No
S116	20	46,XY DSD	Hypomasculinization[d]	15	6.5	9	3	M	Penis bend (twice), hypospadia corrections age 13, 15, and 16	M	M	M	No
S117	23	46,XY DSD	Hypomasculinization[d]	23	6	9	2-3	M	None	F	F→M (age 17)	M	Self-initiated gender change. Relocate to other province and start living as male

Table 7 continued

Patients code	Age at study	Diagnosis	Degree of masculinization at admission					Treatment received prior to study	Gender development history			
			Age	Phallus (cm)	EMS[f]	P/Q[g]	Gender		At birth	Rearing	At study	Gender change[h]
S118	28	46,XY DSD	Hypomasculinization[d]	28	6	9	2-3	M	Chordectomy age 22 (other clinic)	F	F→M (age 17)	M	Self-initiated gender change

Age in years

EMS external masculinization score, *M* male, *F* female, *A* ambiguous gender identity, *F→M* female-to-male gender change, *M→F* male-to-female gender change

[a] 46,XX CAH-SV = congenital adrenal hyperplasia simple virilization type. CYP21 mutation was confirmed in all patients (Juniarto et al., 2014)

[b] 46, XY GD and 46, XX GD = gonadal dysgenesis; subjects had abnormal hormonal testicular function with uni/bilaterally undescended testes. The clinical and biochemical presentation suggest gonadal dysfunction. Serum levels of luteinizing hormone and follicle stimulating hormone were elevated but testosterone, anti-müllerian hormone, and Inhibin are low for age, and no or diminished serum testosterone response to HCG. Androgen action was presumed to be fully effective

[c] 46 XY DSD CAIS / PAIS = complete / partial androgen insensitivity syndromes. A mutation in the AR gene was confirmed (Juniarto et al., 2014)

[d] 46 XY DSD Hypomasculinization refers to hypomasculinization with unknown cause. Molecular and biochemical details are described in Juniarto et al. (2014)

[e] Cloacal exstrophy

[f] Degree of masculinization based on external genital features, ranged from 0 to 12

[g] P/Q = Prader and Quigley stages; stands for degree of genital masculinization in 46, XX (Prader) and 46, XY (Quigley) individuals (Prader, 1954; Quigley et al., 1995)

[h] The term "gender assignment" was applied for the first assigned official gender (usually registration in the birth file). The term "gender reassignment" was applied for a gender change that had been initiated by parents or physician. The term social gender role change was applied for patient who took the initiative to change gender by themselves

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