

Body Image and Self-Esteem in Disorders of Sex Development: A European Multicenter Study

Tim C. van de Grift, Peggy T. Cohen-Kettenis,
Annelou L. C. de Vries,
and Baudewijntje P. C. Kreukels
VU University Medical Center

on behalf of dsd-LIFE

Objective: Disorders/differences of sex development (DSD) refer to congenital conditions with atypical sex development and are associated with psychosexual issues. The aim of this study was to assess body image and self-esteem across the DSD spectrum and to study the impact of diagnosis and mediating characteristics. **Method:** Data collection was part of dsd-LIFE, a cross-sectional study conducted by 14 European expert clinics on wellbeing and health care evaluation of adults diagnosed with DSD. Main outcome measures in the present analyses were the Body Image Scale and Rosenberg Self-Esteem Scale. Additional data were obtained on treatments, openness, body embarrassment, sexual satisfaction, anxiety, and depression. **Results:** The participating sample ($n = 1,040$) included 226 classified as Congenital Adrenal Hyperplasia, 225 as Klinefelter Syndrome, 322 as Turner Syndrome, and 267 as conditions with 46,XY karyotype. Many participants had received hormonal and surgical treatments. Participants scored lower on body image and self-esteem compared to control values, whereas each diagnosis showed different areas of concern. Limited openness, body embarrassment, and sexual issues were frequently reported. Overall body satisfaction was associated with BMI, hormone use, openness, body embarrassment, anxiety, and depression; genital satisfaction with age at diagnosis, openness, sexual satisfaction, and body embarrassment. Body embarrassment, anxiety, and depression predicted lower self-esteem. **Conclusions:** While each DSD showed specific issues related to body image and self-esteem, our findings indicate that the related factors were similar across the conditions. Clinical care on this subject could be improved by giving specific attention to factors like openness, body embarrassment, sexuality, anxiety, and depression.

Keywords: body image, self-esteem, disorders of sex development, intersex, Klinefelter Syndrome, Turner Syndrome

Disorders/differences of sex development (DSD)¹ comprise a group of congenital conditions resulting in an atypical development of sex chromosomes, gonads, and/or genitalia. At a consensus meeting, held in 2005, a classifying framework was introduced

in which the following groups are distinguished: sex chromosomal conditions (including Turner Syndrome [TS] and Klinefelter's Syndrome [KS]), conditions with a 46,XX karyotype (mostly Congenital Adrenal Hyperplasia [CAH]), and conditions with a 46,XY karyotype (including Androgen Insensitivity Syndrome [AIS], Gonadal Dysgenesis [GD], steroid synthesis errors, and [severe] hypospadias) (Hughes, Houk, Ahmed, Lee, & the Lawson Wilkins Pediatric Endocrine Society/European Society for Paediatric Endocrinology Consensus Group, 2006). Despite the presence of different etiological backgrounds and physical characteristics (see Table 1), many of the conditions share impaired gonadal function, atypical sex anatomy, and decreased fertility (Hughes et al., 2006). The consensus statement as well as different support groups emphasize the importance of psychological support as integral part of clinical care (Hughes et al., 2006). Limited comparative and large sample evidence is available on psychological

Tim C. van de Grift, Department of Medical Psychology, Center of Expertise on Gender Dysphoria, and Department of Plastic, Reconstructive & Hand Surgery, VU University Medical Center; Peggy T. Cohen-Kettenis, Department of Medical Psychology, Center of Expertise on Gender Dysphoria, VU University Medical Center; Annelou L. C. de Vries, Department of Child and Adolescent Psychiatry, VU University Medical Center; Baudewijntje P. C. Kreukels, Department of Medical Psychology, Center of Expertise on Gender Dysphoria, VU University Medical Center.

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Correspondence concerning this article should be addressed to Tim C. van de Grift, VU University Medical Center, P.O. Box 7057, 1007 MB Amsterdam, the Netherlands. E-mail: t.vandegrift@vumc.nl

¹ Terminology of intersex or DSD conditions is a sensitive subject, mostly because some terms may be experienced as stigmatizing. To ensure inclusiveness, DSD refers to both disorders and differences of sex development. When applicable, we refer to specific medical diagnoses or symptoms.

Table 1
Overview of the Different Differences/Disorders of Sex Development Diagnoses

Clinical diagnosis	Genetic diagnosis	Gonadal function	Physical characteristics	Predominant gender*	Estimated incidence [†]
Sex chromosomal conditions					
Turner Syndrome	45,X0 karyotype	Ovarian dysgenesis	- Short stature - Turner figure - Hearing problems - Streak ovaries	Female	1:2,500 females
Klinefelter Syndrome	47,XXY karyotype	Testicular dysgenesis	- Small testes, micropenis - Gynaecomastia - Limited body hair - Pear-shaped figure	Male, feel less masculine	1:450–1:600 males
46,XX conditions					
Congenital Adrenal Hyperplasia	21-hydroxylase mutation (CYP21A2)	Excess androgen production	- Clitoris hypertrophy, uro-genital sinus anomaly (C), amenorrhoea (NC) - Virilized puberty - Presence of uterus and ovaries	Female, showing more male-typical behaviour	1:15,000 births
46,XY conditions					
Complete AIS	Androgen receptor mutation	Androgen production, however no sensitivity	- Female external genitalia - Vaginal hypoplasia - Absent uterus and ovaries - Limited body hair	Female	1:40,000 females
Complete GD	Sex determining region mutation on Y-chromosome	Complete ovarian dysgenesis	- Female external genitalia - Presence of vagina and uterus - Streak gonads	Female	1:80,000 females
Other 46XY conditions					
Partial AIS/GD	See above mentioned	Partial variant of above mentioned	Ambiguous genitalia, undervirilized male puberty	Female/male	1:20,000–1:40,000 females
Steroid synthesis errors	5- α reductase, 17- β hydroxylase mutations	Impaired androgen synthesis	Ambiguous/female genitalia, virilized female puberty	Female/male	Unknown, rare
(Severe) hypospadias	Many associated	Mostly typical	Penile anomalies	Male	1:250–1:333 males

Note. AIS = androgen insensitivity syndrome; C = classical congenital adrenal hyperplasia (salt-wasting and simple virilizing types); DSD = disorder/difference of sex development; GD = gonadal dysgenesis; NC = non-classical congenital adrenal hyperplasia.

[†] Incidence rates derived from van Lisdonk (2014), pp. 85–86. * Gender identity and expression are usually less binary than described here.

wellbeing across the different conditions, as well as on the effects of medical interventions and psychosocial characteristics.

Self-perception is defined as awareness of the characteristics that constitute one's self, and includes aspects of body image (cognitions, emotions, and behavior related to one's physique) and self-esteem (ideas and feelings of self-worth; Bem, 1972). A positive body image and self-esteem are important for a person's quality of life, sexuality included. A favorable body image results in less psychiatric issues (Keel & Forney, 2013) and more positive sexual encounters (Woertman & van den Brink, 2012), whereas a higher self-esteem facilitates social participation and enhances feelings of self-efficacy (Judge & Bono, 2001).

Girls and women with TS show a significantly less positive body image compared to controls (Cragg & Lafreniere, 2010; Pavlidis, McCauley, & Sybert, 1995), whereas the body image of women with TS who had previous growth-hormone therapy is more positive (Lagrou et al., 2006). Being overweight, which is more prevalent in women with TS, is strongly associated with a less favorable body image (Cragg & Lafreniere, 2010; Lagrou et

al., 2006). Also, overall self-esteem is lower in women with TS compared to control women (Carel et al., 2006; Fjermestad, Naess, Bahr, & Gravholt, 2016; McCauley, Ross, Kushner, & Cutler, 1995; Pavlidis et al., 1995; Schmidt et al., 2006), as are physique-related self-esteem and social confidence (Cragg & Lafreniere, 2010; McCauley et al., 1995; Pavlidis et al., 1995). Lower self-esteem is associated with higher body mass index (BMI) (Carel et al., 2006; Cragg & Lafreniere, 2010), less sexual experiences (Carel et al., 2006), and more mental health issues (Fjermestad et al., 2016).

Boys and men with KS report less positive body image, and feel less masculine, than controls (Herlihy et al., 2011; Ratcliffe, Bancroft, Axworthy, & McLaren, 1982). In this group, phenotype (e.g., gynaecomastia) and perceived social support are significant predictors of body image (Herlihy et al., 2011). The level of overall self-esteem is, like in women with TS, lower than in controls (Close, Fennoy, Smaldone, & Reame, 2015; Herlihy et al., 2011), and predicted again by phenotype and the experienced quality of social support (Herlihy et al., 2011).

Even though the body image of (mostly operated) women with CAH is moderately positive (Kleinemeier et al., 2010), it is still less positive than in controls (Kuhnle, Bullinger, & Schwarz, 1995; Stikkelbroeck et al., 2003). Women feel less feminine (Kanhere et al., 2015; Kuhnle et al., 1995; Stikkelbroeck et al., 2003), report more sexual avoidance (Kleinemeier et al., 2010; Krege, Walz, Hauffa, Körner, & Rübben, 2000; Kuhnle et al., 1995), and are specifically concerned with their genital appearance and function (Kanhere et al., 2015; Krege et al., 2000; Warne et al., 2005). Their body image is significantly (negatively) related to the degree of virilization at birth (Callens et al., 2012), and less to the physician's outcome scores of surgery (Kanhere et al., 2015; Nordstrom et al., 2010; Stikkelbroeck et al., 2003). Virilization refers to body masculinization due to testosterone, resulting in enlarged clitoris and labial fusion, and consequently frequent genital surgeries that may impact body image as well. Self-esteem is generally moderately positive in women with CAH (de Neve-Enthoven et al., 2016; Kanhere et al., 2015; Warne et al., 2005). However, the scores are generally lower than in controls (Warne et al., 2005).

Women with complete AIS show a rather positive body image (Callens et al., 2014; Wisniewski et al., 2000), with only (absence of) body hair, weight, and genital appearance as areas of concern (Wisniewski et al., 2000). Sexual dysfunction, however, is prevalent (Callens et al., 2012) and related to a negative body image (Callens et al., 2014). A moderately high self-esteem (Callens et al., 2014; de Neve-Enthoven et al., 2016) without significant difference from controls (de Neve-Enthoven et al., 2016) is also reported.

In individuals with ambiguous genitalia (of both XX and XY origin; mostly operated), body satisfaction is relatively positive (Ediati et al., 2015; Migeon et al., 2002; Warne et al., 2005), although less positive than controls (Kleinemeier et al., 2010). Dissatisfaction with genitals, breasts, and (excessive) body hair is reported most frequently (Migeon et al., 2002; Warne et al., 2005). The body image of these women is associated with the level of virilization (and possibly with genital surgeries) and to a lesser extent with physician's scores (Callens et al., 2012). A relatively low self-esteem is reported in this group (Warne et al., 2005). In men with hypospadias, a negative genital perception has been reported in 20% to 80% of boys (Schönbucher, Weber, & Landolt, 2008).

Given the importance of body image and self-esteem as moderators of long-term psychological wellbeing, and the limited number of cross-condition studies in DSD, the present study aims to quantify and compare body image and self-esteem across the spectrum. By assessing these topics across the DSD spectrum for the first time, we hope to further differentiate condition-specific areas of concern and identify shared issues. The latter will be done by studying the impact of medical and psychological characteristics on the outcome, regardless of diagnosis.

Materials and Methods

Procedure

Data collection was part of the dsd-LIFE initiative, a cross-sectional study conducted in 14 European clinics (in Germany, France, the Netherlands, Poland, Sweden, and the United Kingdom) specialized in DSD care and research. The overall objective

was to study the wellbeing and effects of health care of all people of 16 years and older with any of the aforementioned clinically confirmed diagnoses (Röhle et al., 2017). The study was initiated to gain better understanding of the effectiveness of medical interventions on experienced outcome measures, and to translate these findings into recommendations for care and provision of patient information. The study design was developed involving physicians, psychologists, medical ethicists, and support group representatives. In all six countries, the study protocol was approved by ethical committees.²

Between February 2014 and September 2015 all eligible individuals were identified through health records and specialized clinicians, after which people were invited by mail or telephone to participate. In addition, an open call was disseminated through national support groups. A total number of over 3100 eligible people were approached, of whom 1,040 took part in the present study (36%).

After written informed consent, data collection involved digital questionnaires, medical interviewing, physical examination, and/or additional blood and bone tests. Depending on the participant's preferences, study participation would vary from filling out the questionnaires at home and consent to retrieve data from medical files only, to multiple hospital visits for interviewing and additional measurements. After collection, all anonymized diagnostic data were reviewed centrally for accuracy. For an extensive description of the study protocol, readers are referred to Röhle et al. (2017).

Measures

Main outcome measures.

Body Image Scale (BIS). This scale evaluates the satisfaction with 30 body characteristics (Lindgren & Pauly, 1975), rated on a 5-point scale (1 = *very satisfied* to 5 = *very dissatisfied*). Earlier research subdivided the BIS into one overall score and six subscales: social and hair (social refers to socially visible "gendered" body characteristics such as voice), head and neck, muscularity and posture, hip region, breasts, and genital domains (all means) (van de Grift, Cohen-Kettenis et al., 2016). For all scales, a higher mean represents more body dissatisfaction. Confirmatory factor analysis (CFA) confirmed the subscales for both male- ($\chi^2/df = 2.68$) and female-identifying ($\chi^2/df = 1.88$) participants.

Rosenberg Self-Esteem Scale (RSES). This is a widely used measure to assess the level of positive self-evaluation and feelings of self-worth (Rosenberg, 1965). Respondents are asked to rate 10 statements on a scale ranging from *strongly agree* (0) to *strongly disagree* (3). The RSES is reported as one sum score, where a higher value indicates higher overall self-esteem. The RSES

² Institutions that issued ethical approval were as follows: In France: Université Claude Bernard Lyon; Le Centre Hospitalier Universitaire Montpellier; Assistance Publique—Hôpitaux de Paris; Le Centre Hospitalier Universitaire de Toulouse. In Germany: Charité Universitätsmedizin Berlin; Ludwig-Maximilians-Universität, Munich; University of Luebeck; Universitätsmedizin Goettingen; Westfälische Wilhelms-Universität Münster. In Poland: Medical University of Lodz; Children's Memorial Health Institute, Warsaw. Sweden: Karolinska Institutet, Stockholm. In The United Kingdom: University of Birmingham. In the Netherlands: VU University Medical Center, Amsterdam; Radboud University Nijmegen Medical Center, Nijmegen.

showed good psychometric reliability in both genders in the current study (Cronbach's $\alpha_{\text{male}} = .91$, Cronbach's $\alpha_{\text{female}} = .90$).

Other measures.

Questionnaire data. A set of standardized and self-constructed measures was taken from the study participants (Röhle et al., 2017). The current study includes background data on education (ESS Round 6: European Social Survey, 2014; lower/intermediate/higher), household status, and the number of children (self-constructed). Openness about one's condition was rated via agreement with the statement "I can talk freely about my condition" (in general) (self-constructed; 1 [*completely true*] to 5 [*not true at all*]). The overall satisfaction with sex life was measured on a 5-point scale (1 [*very dissatisfied*] to 5 [*very satisfied*]). Participants were also asked whether they felt embarrassed about their body (question from the Coping With DSD scale [Kleinemeier et al., 2010]; 1 [*not at all*] to 5 [*completely*]). To assess the presence of psychological symptoms, the Hospital Anxiety and Depression Scale (subscale scores above 8 indicate clinical issues [Zigmond & Snaith, 1983]) and the (modified) Utrecht Gender Dysphoria Scale were administered (means above 3 [out of 5] were considered to indicate gender dysphoria [Cohen-Kettenis & van Goozen, 1997]). Lastly, people were asked if they ever had been diagnosed with an eating disorder (including anorexia or bulimia).

Medical interview, physical examination, and medical records data. Medical interviewing and physical examination were performed by trained independent study staff using standardized clinical report forms. For the present study, we used interview data collected on age (at the time of diagnosis) and gender identity. Through medical records, information was collected on clinical diagnosis, ambiguity of genitalia at diagnosis (yes/no), past and present hormone therapy (yes/no), and genital surgeries (yes/no). Height and weight were measured to calculate BMI.

Statistics

An analytical plan was developed in collaboration with the statistical support group of the study consortium. For the current study, we grouped participants into four diagnostic categories: women with TS and 46,XX GD; men with KS and 46,XX; women with CAH; and men and women with 46,XY conditions (see Table 1). The first group includes predominantly female-identifying individuals with ovarian dysgenesis and limited surgical interventions; the second group includes predominantly male-identifying individuals with testicular dysfunction, increased prevalence of cognitive issues, and frequent testosterone treatments; the third group includes individuals with CAH only (females with virilized body characteristics); and the last group is more heterogeneous with both female- and male-identifying individuals and more variation in medical treatments, yet all having a 46,XY karyotype. The 5-point scales on satisfaction, support, and sexuality were recoded into scales where a lower score indicated a more positive value (more satisfaction or openness). Group differences on medical and psychosocial characteristics were tested through ANOVA and chi-square tests. A CFA was performed in R statistics to fit the six-factor BIS model, and the reliability of the RSES was assessed through Cronbach's alpha. The BIS output was reported as means, the RSES as sum; both were normally distributed. ANOVA testing was performed to assess differences among the diagnostic groups with regard to the outcome measures. Additional subgroup

comparisons were performed to assess differences within the CAH/46,XY group (based on levels of virilization) and to assess the effects of growth hormones (in TS) and genital surgery (in all). One-sample *t* tests were performed to compare the values of the present sample with control values retrieved from the literature (college and population samples; Sinclair et al., 2010; van de Grift, Kreukels et al., 2016). The association between outcome measures was tested using correlations. To test the influence of predictors of overall and genital body image and self-esteem across the range of DSD diagnoses, linear regressions were performed. The predictors were age at diagnosis, BMI, hormone therapy (yes/no), and genital surgery received (yes/no), openness, sexual satisfaction, body embarrassment, anxiety, and depression. All analyses, except the CFA, were performed in SPSS statistics 22.0.

Results

Background Characteristics

Among the study participants, 226 were diagnosed with CAH (111 salt-wasting, 66 simple virilizing, 34 nonclassical late onset, and 15 other), 225 with KS (204 47,XXY, 6 47,XXY/46,XY, 6 XX males, and 9 other), 322 with TS or 46,XX GD (150 45,XO, 31 45,XO/46,XX, 120 others [variety of deletions, isochromosomes etc.], and 21 with 46,XX GD), and 267 with 46,XY conditions. The last group included XY conditions without androgen action (71 complete AIS and 21 complete GD), partial androgen action (35 partial AIS, 37 partial GD, 45 mixed GD, 18 steroid synthesis conditions, and 5 ovotesticular conditions), hypospadias ($n = 25$), and 46,XY conditions with other/unknown background ($n = 10$). Educational level was normally distributed, with KS participants being least and participants with 46,XY conditions being most educated (see Table 2). The cohort concerned a group of young participants, frequently living without a partner. The KS group was most likely to report to have a partner and child(ren). People in the CAH group had the youngest age at diagnosis (probably due to the highest percentage of ambiguous genitalia at birth or due to an adrenal crisis), TS was frequently diagnosed in childhood or during puberty, 46,XY conditions were commonly diagnosed at a young age or during puberty, and KS was mostly diagnosed during or after puberty. Most participants identified as either male or female (some of whom had changed gender), whereas some identified inter ($n = 7$), other ($n = 3$) or open ($n = 2$).

Medical and Psychological Characteristics

An overview of the characteristics of the study sample is provided in Table 3. In the CAH sample, hormone therapy mostly concerned cortisone, in the KS sample testosterone, in the TS sample growth hormone (past/present use reported by 200) and estrogens, and in the 46,XY sample both estrogen and testosterone therapy. Gonadectomy was mostly performed in the 46,XY group, breast surgery in both the KS (all reductions) and 46,XY groups (six had breast augmentation and 13 had breast reduction), whereas genital surgery was mostly performed in the CAH group (130 had vaginoplasties and 131 had clitoroplasties) and the 46,XY group (50 had vaginoplasties, 37 had clitoroplasties, and 77 hypospadias surgery).

Table 2
Background Characteristics of the Study Sample

Characteristic	Turner syndrome/ 46,XX GD (n = 322)	CAH (n = 226)	46,XY conditions (n = 267)*	Klinefelter syndrome/ 46,XX males† (n = 225)
Mean age (SD)	31.6 (13.1)	30.4 (11.4)	28.8 (12.2)	39.6 (15.1)
Highest education (%)				
Lower	48 (15.9)	40 (18.6)	50 (19.6)	64 (30.2)
Medium	140 (46.4)	104 (48.4)	107 (42.0)	102 (48.1)
Higher	97 (32.1)	52 (24.2)	88 (34.5)	29 (13.7)
Unknown	17 (5.6)	19 (8.8)	10 (3.9)	17 (8.0)
Partner (%)	101 (33.4)	79 (36.4)	87 (33.7)	115 (54.0)
Children (%)	44 (13.7)	38 (16.8)	36 (13.5)	69 (30.7)
Current gender (%)				
Male	0 (—)	5 (2.2)	87 (32.6)	219 (97.3)
Female	322 (100.0)	221 (97.8)	173 (64.8)	1 (4)
Other	0 (—)	0 (—)	7 (2.6)	5 (2.2)
Age at diagnosis (%)				
Below 6 months	47 (16.8)	113 (65.3)	89 (38.2)	12 (6.7)
6 months to 10 years	97 (34.8)	34 (19.7)	44 (18.9)	13 (7.2)
10 to 18 years	110 (39.4)	11 (6.4)	88 (37.8)	47 (26.1)
Above 18 years	25 (9.0)	15 (8.7)	12 (5.2)	108 (60.0)
Ambiguous genitalia at diagnosis (%)	6 (2.0)	166 (79.4)	110 (44.0)	3 (1.5)

Note. CAH = congenital adrenal hyperplasia; GD = gonadal dysgenesis.

† Includes one 47,XXY participant. * Includes 45,XO/46,XY gonadal dysgenesis.

Women with TS were most open about their condition, while those with a 46,XY condition were least open. Participants with 46,XY conditions also showed more sexual dissatisfaction and body embarrassment compared to the other groups. All groups scored relatively high on the anxiety measure, bordering the clinical range. The incidence of clinical gender dysphoria was low (<1%), whereas eating disorders were more prevalent (6%–13%).

Body Image, Self-Esteem, and Diagnosis

Overall, participants had a fairly positive body image (values between “neutral” and “satisfied”; Table 4). Regarding both social

body characteristics (including voice, appearance, and hair growth) and the hip region, participants from the CAH group were least satisfied. Compared to other groups, women diagnosed with TS scored least favorable on head and neck items and on muscularity and posture, although growth hormone use resulted in significantly higher height satisfaction ($M_{growth\ hormone} = 2.77$ (1.1) vs. $M_{no\ growth\ hormone} = 3.18$ (1.1); $t(274) = 3.0$, $p = .003$). Compared with other groups, participants with KS were least satisfied with breasts and genitalia. Within the 46,XY group, participants without androgen action reported higher genital satisfaction ($M = 2.75$ (1.0)) compared to the groups with partial androgen action ($M = 3.15$ (.97)) and with

Table 3
Medical and Psychological Characteristics of the Study Sample

Characteristic	Turner syndrome/ 46,XX GD (n = 322)	CAH (n = 226)	46,XY conditions† (n = 267)	Klinefelter syndrome/ 46,XX males†† (n = 225)	Test statistics
Mean BMI (kg/m ² , SD)	25.1 (5.2)	26.6 (6.2)	24.5 (6.0)	26.0 (5.3)	$F(3, 949) = 5.7$, $p = .001$
Medical care received					
Present hormone therapy (%)	273 (87.2)	217 (96.9)	168 (65.1)	171 (78.8)	$\chi^2(3) = 90.4$, $p < .001$
Surgery received					
Gonadectomy (%)	8 (2.5)	5 (2.2)	184 (69.2)	11 (4.9)	$\chi^2(3) = 512.8$, $p < .001$
Breasts (%)	4 (1.2)	2 (9)	19 (7.1)	14 (6.2)	$\chi^2(3) = 23.1$, $p < .001$
Genitalia (%)	6 (1.9)	136 (59.9)	153 (57.5)	3 (1.3)	$\chi^2(3) = 412.1$, $p < .001$
Support					
Openness about condition (SD)*	2.62 (1.2)	3.34 (1.3)	3.48 (1.4)	3.18 (1.5)	$F(3, 962) = 21.2$, $p < .001$
Sexuality					
Satisfaction with sex life (SD)*	2.83 (1.0)	2.89 (1.2)	3.10 (1.2)	3.02 (1.3)	$F(3, 962) = 2.7$, $p = .05$
Body embarrassment (SD)*	2.44 (1.2)	2.61 (1.2)	2.77 (1.3)	2.62 (1.4)	$F(3, 962) = 3.0$, $p = .03$
Psychological symptoms					
Anxiety (mean HADS, SD)**	6.96 (3.8)	6.95 (4.0)	7.23 (4.1)	6.78 (4.1)	$p > .05$
Depression (mean HADS, SD)**	3.65 (2.9)	4.36 (3.6)	4.15 (3.6)	5.45 (3.9)	$F(3, 980) = 11.5$, $p < .001$
Gender dysphoria (%)	4 (1.2)	1 (4)	2 (8)	2 (9)	$p > .05$
Eating disorder (%)	40 (13.3)	28 (13.0)	28 (10.9)	13 (6.2)	$p > .05$

Note. BMI = body mass index; CAH = congenital adrenal hyperplasia; GD = gonadal dysgenesis; HADS = Hospital Anxiety and Depression Scale. † Includes 45,XO/46,XY gonadal dysgenesis. †† Includes one 47,XXY participant. * 5-point scale; for all marked scales a lower score indicates a more positive value. ** 0 = minimal symptoms to 21 = maximal, scores of ≥ 8 indicate clinical anxiety/depression.

Table 4
Body Image and Self-Esteem Scores

Outcome	Turner syndrome/ 46,XX GD (n = 322)	CAH (n = 226)	46,XY conditions (n = 267)	Klinefelter syndrome/ 46,XX males (n = 225)	Test statistics
Body image (BIS, SD)*	2.61 (.66)	2.55 (.70)	2.48 (.69)	2.56 (.66)	$p > .05$
Social and hair growth (SD)	2.58 (.73)	2.69 (.87)	2.42 (.80)	2.47 (.71)	$F(3, 951) = 5.3, p = .001$
Head and neck (SD)	2.29 (.69)	2.22 (.83)	2.16 (.68)	2.19 (.65)	$p > .05$
Muscularity and posture (SD)	2.61 (.72)	2.35 (.73)	2.37 (.69)	2.39 (.73)	$F(3, 954) = 8.1, p < .001$
Hip region (SD)	2.73 (.86)	2.83 (.88)	2.57 (.93)	2.62 (.80)	$F(3, 955) = 3.8, p = .01$
Breasts (SD)	2.74 (.94)	2.65 (1.1)	2.61 (1.0)	2.88 (1.1)	$F(3, 938) = 2.9, p = .03$
Genitals (SD)	2.79 (.76)	2.59 (.90)	2.98 (1.0)	3.31 (1.0)	$F(3, 946) = 22.6, p < .001$
Self-esteem (RSES, SD)**	19.1 (5.6)	20.7 (6.1)	19.7 (6.1)	19.5 (6.1)	$F(3, 978) = 2.9, p = .04$

Note. BIS = Body Image Scale; CAH = congenital adrenal hyperplasia; GD = gonadal dysgenesis; RSES = Rosenberg Self-Esteem Scale; norm values BIS overall male = 2.15, female = 2.38; BIS social and hair male = 2.26, female = 2.38; BIS head and neck male = 2.04, female = 2.31; BIS muscularity and posture male = 2.16, female = 2.35; BIS hip region male = 2.17, female = 2.55; BIS breasts male = 2.31, female = 2.28; BIS genitals male = 1.95, female = 2.27; RSES male = 22.4, female = 22.8.

* 1 = very satisfied to 5 = very dissatisfied. ** 0 = lowest self-esteem to 30 = highest self-esteem.

hypospadias ($M = 3.29 (1.1)$; $F(2, 249) = 5.8, p = .004$). Such difference was not found in other body image domains. Compared to male and female control values available in the literature (van de Grift, Kreukels et al., 2016), study participants reported significantly higher body dissatisfaction, most strongly on the genital subscale ($M_{male\ control} = 1.95$; $t(949) = 30.4, p < .001$ and $M_{female\ control} = 2.27$; $t(949) = 20.2, p < .001$).

All group means on self-esteem fell within the normal range (15 to 25), but had high standard deviations (see Table 4). The level of self-esteem was highest in the CAH group and lowest in the TS group. No significant effect of growth hormone use on self-esteem was found within the latter group. Study participants scored significantly lower on self-esteem than control males ($M = 22.4$; $t(981) = -14.2, p < .001$) and females ($M = 22.8$ [Sinclair et al., 2010]; $t(981) = -16.1, p < .001$). Overall body dissatisfaction was on the one hand associated with genital

body dissatisfaction ($r(949) = .53, p < .001$) and on the other hand with overall self-esteem ($r(961) = -.47, p < .001$).

Predictors of Body Image and Self-Esteem

Table 5 displays the results and model statistics of the linear regressions on (overall and genital) body image and self-esteem over the sample as a whole. Positive overall body image was associated with a lower BMI, absence of hormone therapy, more openness about one's condition, low body embarrassment, and low anxiety and depression scores. Associations of a more positive genital body image included a younger age at diagnosis, more openness about one's condition, higher sexual satisfaction, and a lower degree of body embarrassment. Finally, higher self-esteem was associated with low levels of body embarrassment, anxiety, and depression.

Table 5
Predictors of (Genital) Body Image and Self-Esteem Across the Sample

Predictor	Overall body image (BIS)		Genital body image (BIS)		Self-esteem (RSES)	
	β	p -value	β	p -value	β	p -value
Age at diagnosis	.02	.50	.13	<.001	.00	1.0
BMI	.15	<.001	.05	.13	.03	.27
Present hormone therapy	.10	.001	.03	.39	-.02	.54
Genital surgery received	-.05	.18	-.04	.29	.06	.05
Openness about condition	.07	.02	.12	<.001	-.05	.08
Sexuality						
Satisfaction with sex life	.07	.05	.14	<.001	-.03	.29
Body embarrassment	.31	<.001	.24	<.001	-.19	<.001
Psychological symptoms						
Anxiety	.13	.001	.06	.13	-.28	<.001
Depression	.11	.005	.11	.01	-.34	<.001

Note. Linear regression of medical and psychosocial predictors of overall and genital body image, and self-esteem. Higher BIS scores correspond with more dissatisfaction, whereas a higher RSES score corresponds with higher overall self-esteem. The variable labels on received treatment are 1 for confirming and 0 for not-confirming, for the satisfaction, sexuality, and psychological symptoms variables; a lower score implies a more positive score. Overall body satisfaction model: adjusted $R^2 = .303, F(9, 716) = 34.6, p < .001$; genital satisfaction model: adjusted $R^2 = .238, F(9, 706) = 24.5, p < .001$; self-esteem model: Adjusted $R^2 = .450, F(9, 721) = 65.5, p < .001$. BIS = Body Image Scale; BMI = body mass index; RSES = Rosenberg Self-Esteem Scale.

Discussion

The joint effort of multiple European expert clinics enabled the study of body image and self-esteem across different DSD conditions, including a variety of medical and psychosocial predictors. Although body image and self-esteem were fairly positive, participants scored significantly unfavorable compared to the available control values.

Participants diagnosed with CAH showed highest dissatisfaction with social characteristics (such as voice) and hair growth, most likely due to increased virilization. Virilization was earlier described as an influential factor on body image (Callens et al., 2012). Another area of concern was the hip region, which may be related to the highest average BMI compared to other included groups. Prior research on body composition of women with CAH indicated increased waist–hip ratios, suggesting a less gynoid body shape (Falhammar et al., 2007). Surprisingly, and in contrast with other studies (Kanhere et al., 2015; Kregel et al., 2000; Warne et al., 2005), genital body satisfaction was relatively high in the CAH group. The majority of this group had (some degree of) ambiguous genitalia at birth and received genital surgeries for that reason. The finding that this group was relatively satisfied with their genital surgeries is likely to have contributed to the genital satisfaction rates. However, this finding should be interpreted prudently as there was no control group without surgery with which to compare present results. Yet male and female controls in previous studies still showed significantly higher degrees of genital satisfaction, a finding reported in other studies as well (Kleinemeier et al., 2010; Kuhnle et al., 1995; Stikkelbroeck et al., 2003). Lastly, the suboptimal rates of sexual satisfaction and body embarrassment illustrate that genital satisfaction remains a subject of attention in psychological health care for women diagnosed with CAH.

In relation to the other subgroups, participants diagnosed with TS reported highest dissatisfaction with head and neck, as well as with muscularity and posture. This shows that the associated syndromic physical characteristics, such as webbed neck and short stature, translate to areas of discomforted body image. The more positive findings with regard to body image in the 200 participants who received growth hormones compared with those who did not receive this treatment confirm earlier findings (Lagrou et al., 2006). However, clinicians should be aware of the limitations of this treatment; participants with TS reported the least positive overall body image compared with the other DSD groups, and growth hormones did not improve self-esteem. As suggested earlier, this may be related to the higher BMI values (in relation to controls; Cragg & Lafreniere, 2010; Lagrou et al., 2006) or that end height is still relatively low. Also, TS with its physical attributes expanding beyond sex characteristics only may be the most visible of all conditions studied here, possibly increasing the likelihood of negative impact of one's appearance on life.

Not surprisingly, the KS group reported highest dissatisfaction on breasts and genitals. The syndrome comes with high incidences of gynaecomastia and low testicular volume, which impact body image (Herlihy et al., 2011). Mastectomy or testicular implant surgery were rare in our sample, and many participants with KS were diagnosed at a later age, delaying supportive care. Also, the KS group predominantly included male-identifying participants, who may have felt they had had to meet the masculine ideal, which

may be problematic and resulting in body image difficulties (Ratcliffe et al., 1982).

Participants with an 46,XY condition showed—not unexpectedly, given the heterogeneous group composition—divergent results with regard to body image; female-identifying participants with no androgen action showed more positive scores than participants with partial androgen action or hypospadias. This confirms earlier research that the level of virilization is associated with body image problems (Callens et al., 2012), and that the body image of people with (previously) ambiguous genitalia and hypospadias is frequently a matter of concern (Kleinemeier et al., 2010; Schönbacher et al., 2008). Unsurprisingly, the highest body dissatisfaction rate was reported on genitalia. The groups with partial androgen action and hypospadias indeed had most genital surgeries, which they were least satisfied with, compared to the other DSD groups, which again may have contributed to this high genital dissatisfaction rate. Further complicating factors in the development of a positive self-image are the limited openness, the embarrassment, and low sexual satisfaction found in this group.

Most studies found that people with DSD reported self-esteem scores lower than control values (Carel et al., 2006; Ee et al., 2015; Fjermestad et al., 2016; Herlihy et al., 2011; McCauley et al., 1995; Pavlidis et al., 1995; Schmidt et al., 2006; Warne et al., 2005), an outcome that is consistent with our findings. The present study, however, was able to confirm this finding in a large sample, and showed that similar values were obtained throughout the different diagnostic groups. This emphasizes that besides physical factors like phenotype (Herlihy et al., 2011; Warne et al., 2005) and BMI (Carel et al., 2006; Cragg & Lafreniere, 2010), shared experiences and psychosexual concerns across the DSD spectrum may result in body image and self-esteem issues of similar degree.

The importance of recognizing these shared experiences across the diagnostic groups was further emphasized by the identification of predictors of body image and self-esteem. As mentioned earlier, a higher BMI was associated with less body satisfaction. Hormone use was also associated with body dissatisfaction, possibly indicating that participants who did not need hormone therapy were more positive about their bodies (although a reverse relationship may be the case as well) and experienced less physical side effects due to the artificial hormones (e.g., estrogen-related weight gain). Additional associations of unfavorable body image were symptoms of anxiety and depression. Especially, anxiety levels were high across the subgroups, bordering clinical cut-offs. Possible explanations of increased anxiety levels include identity-related distress, lack of openness, relatively low numbers of participants having a partner or children, side effects of hormone therapy, and/or negative social experiences. Openness had a positive influence on overall and genital body image. Past medical protocols have been hesitant to advise people diagnosed with DSD conditions to be open (Lossie & Green, 2015). People were advised to be secretive in order to prevent psychological harm, while the present findings suggest that more openness supports positive body image and self-esteem. Being open about your condition may reduce stigma, support positive (sexual) experiences, and provide a sense of control. Openness applies to both the clinical context as well as to daily life. Support group contact can be of

importance as it can create a (first) safe space to be open about one's DSD condition. Clinicians best cooperate with support groups by referring to one another, by jointly developing information material and the early signaling of structural barriers to health care. Genital body image was strongly associated with sexual satisfaction, a finding consistent with earlier research (Callens et al., 2014). Genital self-image is closely related to the quality of sex life through genital functioning, sexual avoidance, and experienced (self-)stigma (Woertman & van den Brink, 2012). Lastly, lower self-esteem was mostly associated with other psychological measures, including body embarrassment, anxiety, and depression. As all these measures were high—compared to norm values—throughout the sample, clinicians should be attentive to these psychological issues in these groups. Given the complex interrelationship between biological and psychosocial factors in DSD and the possible negative psychological outcomes of past psychosocial support of suboptimal quality (e.g., by promoting secrecy [Lossie & Green, 2015], or focusing on outcomes less relevant for the group themselves [Liao & Roen, 2014]), investment in and training of specialized mental health professionals for interdisciplinary treatment facilities is warranted.³

The present study was limited in some aspects. The first limitation was the representativeness of the sample. People with negative health care experiences may be more likely to be lost to follow-up and may not have participated. Unfortunately, due to the recruitment strategy (including open calls via support groups), it was impossible to reliably compare characteristics of nonparticipants with participant data. Also, on several important variables (e.g., the role of early genital surgery), the data remained inconclusive due to the absence of control groups (control values from the literature only). The study was potentially limited by psychometric biases, such as recall and confirmation bias, and potential false associations between instruments resulting from similar answering regardless of the measured subject. Also, we are aware that the present subdivision of diagnostic groups and medical interventions does not do justice to the full complexity of all diagnoses, medical treatments, and health care practices. It is therefore important to further study body image and self-esteem in differentiated DSD subgroups. This should include studies using validated measures, as well as explorative hypotheses-generating studies on body image, sexuality, and their relationship. Also, we encourage more research studying the effects of social determinants such as social support, discrimination, minority stress, and trust in medical professionals on these outcome measures.

Conclusion

The present study highlights the significant psychological issues related to body image and self-esteem of people with DSD. While some reported fairly positive values, each diagnostic group showed specific areas of concern. These concerns were often related to body composition (e.g., stature or BMI) and sex-atypical physical characteristics. Importantly, it appeared to be relevant to look at shared experiences across the DSD spectrum; factors such as sexual satisfaction, openness and body embarrassment, anxiety and depression were significantly associated with body image and self-esteem throughout the sample. Therefore, it is relevant to

design affirmative services for adults that address these topics, promote openness, and reduce stigma whenever possible. Interdisciplinary services—in cooperation with support groups—can integrate biological, psychological, and social support in order to improve overall psychosexual wellbeing of individuals with DSD. For those with severe body image and self-esteem issues, existing psychotherapeutic treatments (e.g., focusing on body acceptance or dysfunctional cognitions/behaviors) are best tailored to the specific needs of this group.

³ Multiple online resources are available for (non)specialized mental health professionals, including the APA factsheet (<https://www.apa.org/topics/lgbt/intersex.pdf>), websites of support groups (e.g., <http://www.accordalliance.org/>), information by this study consortium (<http://www.dsd-life.eu/>), and private initiatives (e.g., <http://www.dsdteens.org/> and <http://www.dsd-families.org/>).

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