

# Emotional and Behavioral Problems in Late-Identified Indonesian Patients with Disorders of Sex Development

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## Emotional and behavioral problems in late-identified Indonesian patients with disorders of sex development



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### ABSTRACT

**Objective:** The aim of this study is to investigate emotional and behavioral problems among Indonesian patients with disorders of sex development (DSD) who recently came under clinical management. As diagnostic procedures and treatment had been delayed, patients progressively developed ambiguous bodies, difficult to conceal from outsiders.

**Method:** We compared 118 Indonesian patients with DSD aged 6–41 years (60 children, 24 adolescents, 34 adults) and 118 healthy control subjects matched for age, gender, and residential settings. We used the Child Behavioral Checklist (CBCL), Youth Self-Report (YSR), and Adult Self-Report (ASR) to examine differences between patient and control groups as well as differences within patients groups.

**Results:** On the CBCL, parents of young children with DSD reported significantly more emotional and behavioral problems than parents of matched control. Parents of daughters with CAH reported that their daughters withdrew themselves from social interactions. On the ASR, adults with DSD reported significantly more internalizing problems than controls, particularly anxiety and depression. No other differences in emotional functioning were found across different diagnostic groups.

**Conclusions:** Indonesian patients with DSD who were untreated for most of their lives suffered more emotional and behavioral problems than matched controls. Differences and similarities between our findings and observations in patients from Western countries will be discussed.

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### Introduction

Disorders of sex development (DSD) refer to a group of congenital conditions in which the development of chromosomal, gonadal, and anatomical sex is atypical [1]. The atypical sex development starts prenatally and leads to the development of ambiguous internal and/or external genitalia, with development of ambiguous secondary sex characteristics (e.g. development of small breasts and facial hair) at puberty. The large majority of patients are infertile.

The combination of an atypical genital and body appearance and (suspected) infertility puts patients in a delicate position in society. In Western countries, patients will be referred for diagnostic procedures and medical treatment soon after identification of their atypical sex development. Diagnostic evaluation will be performed, and patients and

their parents will be informed about their condition and available treatments. In addition, hospital staff and patient support groups can provide emotional support and advice with respect to communication about their condition within their families and communities. Some of the medical interventions are necessary for survival (e.g. glucocorticoid and aldosterone replacement in CAH). Among the treatments not necessary for survival are some surgeries directed to correct atypical genital appearance and prevent atypical pubertal progression. A rationale for this practice is protection against social stigmatization and providing optimal opportunities for social participation [2,3]. However, this practice is currently under debate.

In Indonesia, DSD is largely unknown, even among health practitioners. As a consequence, DSD is often identified late and diagnostic procedures are postponed or not performed. The large majority of the patients who presented at our clinic had never undergone diagnostic evaluation or received medical treatments. Many of these patients lived with ambiguous genitalia and were developing (adolescents) or had developed (adults) bodies with both male and female secondary sex characteristics. Some patients doubted their gender as did their parents and community members. Many patients reported exclusion and other signs

**Abbreviations:** DSD, disorders of sex development; CBCL, Child Behavioral Checklist; YSR, Youth Self Report; ASR, Adult Self Report.

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of social stigmatization due to their physical appearance. We hypothesized that body ambiguity was stressful and could lead to the development of emotional problems. Our study aimed to investigate self-reported emotional problems in children, adolescents, and adult patients with DSD in Indonesia.

## Method

### Study design

In this study, we investigated patient- or parent-reported emotional and behavioral problems in Indonesian children, adolescents, and adults with a disorder of sex development (DSD). Findings in patients were compared to findings in matched control subjects. All patients and parents consulted the DSD team of the Dr. Kariadi Hospital and Faculty of Medicine of the Diponegoro University (FMDU), Indonesia. The study protocol was approved by the board of the ethical committee from FMDU.

### Participants

#### Patients with DSD

All patients with a proven diagnosis of DSD who were under clinical management of the DSD Team of the Dr. Kariadi Hospital and FMDU [4] were invited for study participation. Patients under age 6, patients with intellectual disabilities (indicated from parent reports on their child's academic achievements and/or observed by the medical doctor in interaction with the patient), patients with a genital anomaly and features suggestive of malformation syndromes [5] and patients with sex chromosome DSD without mosaicism were excluded for study participation. We applied these exclusion criteria as a reliable assessment of emotional and behavioral problems is difficult in preschool children and patients with limited intellectual capacities. Patients with malformation syndromes and Turner syndrome often suffer from additional somatic pathology. Low IQ and intellectual disabilities are more frequently seen in patients with malformation syndromes and patients with Turner and Klinefelter syndromes. The additional somatic pathology may also be related to (other types of) emotional problems. In patients with Turner and Klinefelter syndromes specific psychopathology is more frequently seen compared to other groups of patients with DSD [6,7]. 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 participation. The response rate was 70.2%. No specific characteristics were found among parents and patients who declined participation. Thirty-five adults, 24 adolescents, 23 parents of adolescents and 60 parents of young children filled out the questionnaires. Table 1 summarizes patients and diagnoses. Details of the individual clinical data were presented in Appendix A.

**Table 1**  
Clinical diagnosis of patients in the study.

DSD diagnosis	Age			Total
	6–11	12–17	18+	
Sex chromosome DSD				
46, XY DSD	Mosaics	3	0	3
	AIS <sup>a</sup>	5	5	6
	Gonadal dysgenesis <sup>b</sup>	9	7	12
	Hypomasculinization e.c.i.	25	9	7
46, XX DSD	CAH – SV <sup>c</sup>	18	2	4
	Gonadal dysgenesis	0	0	1
	Cloacal malformation	0	1	1
Total		60	24	34
				118

Note. CAH-SV = simple virilizing type of congenital adrenal hyperplasia.

<sup>a</sup> AR gene mutation was confirmed [4].

<sup>b</sup> A condition in which patients had abnormal hormonal testicular function with uni/bilaterally undescended testes. The clinical and biochemical presentations suggest gonadal dysfunction [4].

<sup>c</sup> CYP 21 mutation was confirmed [4].

### Control group

For each participating patient, a healthy control subject was found matched for age, gender, and residential settings. Residential setting (rural, suburban, or urban area) was chosen as matching criteria in order to compare subjects living under comparable socioeconomic conditions. Control subjects were approached by local leaders (in Bahasa: Pak RT or Pak Lurah) or midwives. After a potential matched control subject was identified, an invitation to join the study was given. In order to guarantee the privacy of the patients, the Pak RT or Pak Lurah and the matched control subjects were informed that this study was a population study on emotional and behavioral problems carried out at the Faculty of Psychology, Diponegoro University. None of the potential matched control subjects declined participation in the study. After the control subjects gave their written consent to participate in the study, data collection was conducted following the similar procedure as the patients with DSD.

### Measures

Data on emotional and behavioral problems were obtained using ASEBA (Achenbach System of Empirically Based Assessment) scales: the Child Behavior Checklist/CBCL 6–18 [8], the Youth Self-Report/YSR [8], and the Adult Self-Report/ASR [9]. The ASEBA scales are widely applied as a screening instrument for psychopathology [8, 9]. Cut-off scores help to identify patients who score in borderline and clinical ranges and are at risk to develop severe emotional and behavioral problems. These measures assess behavioral and emotional problems reported over the past six months. Each item is rated on a 3-point scale: 0 (not true), 1 (somewhat or sometimes true), and 2 (very true or often true). Higher scores indicate a higher level of emotional and behavior problems.

#### CBCL/6–18

The CBCL/6–18 is a 120-item standardized parent-report measure for emotional and behavioral problems in children aged 6 to 18 years [8]. It measures on eight scales: Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behavior, and Aggressive Behavior. This measure had been validated across 30 societies and has satisfactory reliability and psychometric quality for assessing problem behavior in children across cultures [10]. The validation of the Indonesian version of the CBCL/6–18 was obtained from a study involving 107 parents of children aged 6–18 in the Central Java province [11]. The Cronbach's alphas found on scales of the Indonesian version of the CBCL/6–18 ranged between 0.56 (Social problems) and 0.94 (Total Problems) [11]. These Cronbach's alphas of the Indonesian CBCL/6–18 are comparable to Cronbach alphas found in other studies [12].

#### YSR

The YSR is a 119-item standardized self-report measure for emotional and behavioral problems in youth aged 11 to 18 years [8]. It includes eight scales similar to the CBCL. The YSR had been validated across 23 countries, and had satisfactory psychometric quality for assessing problem behavior among adolescents across different cultures [13]. The validation of the Indonesian version of the YSR was obtained from a study involving 1154 high school students in Central Java province. Cronbach's alphas of the Indonesian version of the YSR ranged between 0.62 (Social Problems) and 0.92 (Total Problems) [11]. To assess the factor structure of the Indonesian translation of YSR, we followed procedures for confirmatory factor analysis described by [13] and found comparable results. The RMSEA of 0.03 was within the range of previous report [13] and indicated good fit. The CFI and TLI were 0.86 and 0.85 indicating acceptable fit [13]. The factor loadings ranged from 0.28 to 0.77 with median factor loading 0.57, whereas the factor covariance ranged from 0.45 to 0.98 with median covariance of 0.73. Thus, the results



confirmed that the eight structure model also holds for the Indonesian translation of YSR [11].

#### ASR

The ASR is a 131-item standardized self-report measure of emotional and behavioral functioning in adults aged 18 or older [9]. It comprised eight scales: Anxious/Depressed, Withdrawn, Somatic Complaints, Thought Problems, Attention Problems, Aggressive Behavior, Rule-Breaking Behavior, and Intrusiveness. The validity and reliability of the Indonesian translation of the ASR were assessed following procedures described by Achenbach and Rescorla [9]. The data was obtained from a study involving 1091 university students in the Central Java province. Cronbach's alphas ranged from 0.59 (Thought Problems) to 0.94 (Total Problems). The RMSEA of 0.02 was within the range of Achenbach and Rescorla [9] and indicated good fit. The CFI and TLI were both 0.90 indicating a good fit [10]. The factor loadings ranged from 0.17 to 0.78 with a median factor loading of 0.61, whereas the factor covariance ranged from 0.34 to 0.89 with a median covariance of 0.63. The results confirmed that the eight-structure model also holds for the Indonesian translation of ASR [11].

#### Socio-demographic characteristics

In addition to data on emotional and behavioral problems, we also collected data on socio-economic status, ethnic and cultural background, including age, gender, residence, ethnicity, religion, education (the highest level attained), and occupation.

#### Procedures

Data collection was carried out between March 2007 and May 2011. After a DSD was diagnosed [4], patients were invited to participate in this study. Oral and written study information was provided by a medical doctor (AZJ). After participants had given their informed consent, an appointment was made for the study. For participants under age 18, informed consent was obtained from the parents. The psychological assessment was conducted in the hospital or at home by a trained psychologist (AE). In participants with limited educational background (i.e. illiterate) or participants unfamiliar with self-report, ASEBA questionnaires were administered orally by the psychologist (AE). Similar to the paper-pencil version, participants had to indicate their response (0–1–2) for each question, the psychologist ticked off their answer in the scoring sheet.

#### Statistical analysis

We used the paired Wilcoxon sign-rank test to test for differences in levels of emotional and behavioral problems between patient and matched control groups and differences between these groups separate for males and females. We also dichotomized the scores on the ASEBA scales into two categories: individuals scoring above the borderline cut-off point and individuals scoring below the borderline cut-off point [8,9]. This classification distinguishes between severe emotional and behavioral problems/psychopathology and problems in the normal range. With this classification we tested whether there were differences between age groups and between diagnostic groups in terms of the number of individuals scoring above the cut-off point using Fisher's Exact Test. We also used the dichotomized scores to test differences in the number of individuals scoring above the cut-off point between patient and matched control groups in which the age groups were combined. For this analysis we used the McNemar test. Differences were considered significant at  $p < 0.05$  (two-sided).

## Results

#### Participant characteristics

As shown in Table 2, patients and matched controls did not differ with respect to their socio-demographic, ethnic and cultural background. The majority of participants was male, lived in rural areas, came from the Central Java province, and was Javanese and Moslem. The parents' educational background varied from illiterate to university level, and the majority had attended high school. There was a difference between groups with respect to occupation; more parents of patients worked in the lower-income sector or were unemployed.

#### Emotional and behavioral problems in patient and control groups

In Table 3, sections A to D, the results on reported emotional and behavioral problems in patient and control groups are presented.

#### Children

On the CBCL, parents of children aged 6–11 with DSD reported more emotional and behavioral problems in their children than parents of the matched control children (Total Problems:  $p = 0.02$ ). More specifically parents reported Externalizing Problems ( $p = 0.03$ ); Social Problems ( $p = 0.04$ ), Attention Problems ( $p = 0.03$ ) and Aggressive Behavior ( $p = 0.02$ ). No significant differences were found for the other scales. A separate analysis by gender revealed that these emotional problems were particularly reported by parents of boys (Total Problems:  $p = 0.05$ ; Externalizing Problems  $p = 0.04$  and Aggressive Behavior ( $p = 0.02$ ). Emotional problems reported by parents of girls with DSD did not differ from controls (see Table 3A).

#### Adolescents

No differences on the CBCL and YSR scores were found between the adolescent patients and the matched control groups (see Table 3B and C).

#### Adults

As reported in Table 3D, adults with DSD reported more Internalizing Problems than matched control adults did ( $p = 0.04$ ); more specifically they reported higher scores on the Anxious/Depressed scale ( $p = 0.01$ ). Particularly men with DSD gave higher ratings on the Anxious/Depressed scale than the matched control men did ( $p = 0.04$ ), whereas women with DSD more often reported social isolation compared to their matched controls ( $p = 0.02$ ).

**Table 2**  
On demographic and cultural characteristics.

Characteristics background	Patients with DSD (n = 118)	Matched controls (n = 118)	p-Value
Age of study	13.8 ± 7.4	14.2 ± 7.1	0.69
Region			
Central Java province	100 (84.7)	108 (91.5)	0.12
Other provinces in Java	12 (10.2)	9 (7.6)	
Outside Java island	6 (5.1)	1 (.8)	
Ethnicity			
Javanese	108 (91.5)	106 (89.8)	0.82
Non Javanese	10 (8.5)	12 (10.2)	
Religion			
Islam	112 (94.9)	108 (91.5)	0.44
Non Islam	6 (5.1)	10 (8.5)	
Education—Father*	n = 116	n = 114	
Illiterate	18 (15.5)	15 (13.2)	0.62
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*	n = 116	n = 117	
Illiterate	22 (19.0)	14 (12.0)	0.33
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*	n = 116	n = 114	
Unemployed	6 (5.2)	5 (4.4)	0.06
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*	n = 116	n = 117	
Unemployed	57 (49.1)	39 (33.3)	0.02
Labor	32 (27.6)	35 (29.9)	
Self-employed	12 (10.3)	28 (23.9)	
Staff/Office job	15 (12.9)	15 (12.8)	

Note. Data presented in n (%), except age: in median (range). Fisher's exact test was applied; significant at  $p < 0.05$ . \* indicates differences in n.

**Table 3**  
Emotional and behavioral problems reported by parents, adolescents and adults from patient and control groups.

A. Parental report on emotional and behavioral problems in children aged 6–11									
CBCL6–18 scales <sup>a</sup>	Study groups		p-Value	Boys		p-Value	Girls		p-Value
	DSD	Controls		DSD	Controls		DSD	Controls	
	n = 60	n = 60		n = 42	n = 42		n = 18	n = 18	
	Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)	
Total problems	23.5 (5–59)	13 (2–69)	0.02	22 (5–59)	13 (2–57)	0.05	25 (9–56)	15.5 (3–69)	0.14
Internalizing problems	4 (0–18)	3 (0–25)	0.45	3 (1–17)	3 (0–25)	0.87	5.5 (1–19)	4 (0–21)	0.48
Externalizing problems	7.5 (0–35)	5 (0–22)	0.03	7.5 (1–31)	5 (0–22)	0.04	6.5 (1–35)	5.5 (0–19)	0.48
<i>Scales:</i>									
Anxious/depressed	1 (0–8)	1 (0–10)	0.95	1 (0–8)	1 (0–10)	0.98	1.5 (0–7)	1.5 (0–10)	0.85
Withdrawn/depressed	1.5 (0–12)	1 (0–7)	0.16	1 (0–12)	1 (0–7)	0.67	2 (0–11)	1.5 (0–6)	0.11
Somatic complaints	1 (0–5)	0 (0–8)	0.61	0 (0–4)	0 (0–8)	0.63	1 (0–5)	1 (0–8)	0.89
Social problems	3.5 (0–10)	2 (0–11)	0.04	3 (0–10)	2 (0–11)	0.08	1 (0–8)	4 (1–8)	0.32
Thought problems	1 (0–6)	0 (0–8)	0.91	1 (0–6)	0 (0–7)	0.48	0 (0–4)	0.5 (0–8)	0.38
Attention problems	2 (0–14)	1 (0–11)	0.03	2 (0–14)	1 (0–11)	0.16	3 (0–8)	0.5 (0–11)	0.15
Rule-breaking behavior	2 (0–13)	2 (0–8)	0.23	2 (0–13)	2 (0–8)	0.11	1.5 (0–11)	2 (0–5)	0.70
Aggressive behavior	5.5 (0–24)	3 (0–25)	0.02	5.5 (0–20)	3 (0–18)	0.02	5.5 (1–24)	4.5 (0–14)	0.42
B. Parental report on emotional and behavioral problems in adolescents aged 12–17									
CBCL6–18 scales <sup>a</sup>	Study groups		p-Value	Boys		p-Value	Girls		p-Value
	DSD	Controls		DSD	Controls		DSD	Controls	
	n = 23 <sup>b</sup>	n = 23		n = 15	n = 15		n = 8	n = 8	
	Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)	
Total problems	9 (4–29)	10 (0–43)	0.40	8 (4–29)	10 (1–43)	0.86	17.5 (5–21)	10.5 (0–21)	0.31
Internalizing problems	3 (0–14)	4 (0–14)	0.59	2 (0–11)	4 (0–14)	0.81	5 (1–14)	3 (0–13)	0.16
Externalizing problems	2 (0–10)	1 (0–12)	0.69	2 (0–10)	1 (0–12)	0.93	1 (0–8)	2.5 (0–6)	0.57
<i>Scales:</i>									
Anxious/depressed	1 (0–4)	1 (0–5)	0.73	1 (0–4)	1 (0–5)	0.66	1 (0–2)	0.5 (0–3)	0.91
Withdrawn/depressed	2 (0–11)	1 (0–10)	0.68	1 (0–7)	2 (0–6)	0.55	3 (0–11)	1 (0–10)	0.19
Somatic complaints	0 (0–6)	1 (0–7)	0.60	0 (0–3)	0 (0–7)	0.95	1.5 (0–6)	0.5 (0–4)	0.41
Social problems	2 (0–7)	1 (0–7)	0.63	2 (0–7)	1 (0–7)	0.52	2 (0–5)	1.5 (0–5)	0.99
Thought problems	0 (0–3)	0 (0–4)	0.99	0 (0–3)	0 (0–4)	0.78	0.5 (0–2)	0 (0–2)	0.81
Attention problems	2 (0–6)	1 (0–6)	0.99	1 (0–5)	1 (0–5)	0.40	2.5 (0–6)	0.5 (0–6)	0.37
Rule-breaking behavior	0 (0–5)	0 (0–4)	0.36	0 (0–5)	1 (0–4)	0.51	0.5 (0–2)	0 (0–3)	0.75
Aggressive behavior	1 (0–8)	1 (0–8)	0.82	1 (0–8)	1 (0–8)	0.99	0.5 (0–7)	1.5 (0–4)	0.80
C. Self-report on emotional and behavioral problems in adolescents aged 12–17									
YSR 12–17 scales <sup>a</sup>	Study groups		p-Value	Boys		p-Value	Girls		p-Value
	DSD	Controls		DSD	Controls		DSD	Controls	
	n = 24	n = 24		n = 15	n = 15		n = 9	n = 9	
	Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)	
Total problems	18.5 (3–65)	18.5 (0–107)	0.86	14 (3–52)	19 (0–50)	0.57	28 (7–60)	18 (3–96)	0.50
Internalizing problems	5 (0–25)	5.5 (0–35)	0.51	3 (0–25)	6 (0–16)	0.27	11 (1–25)	5 (0–35)	0.12
Externalizing problems	5 (0–14)	5.5 (0–22)	0.38	3 (1–16)	6 (0–15)	0.53	6 (0–21)	5 (1–21)	0.89
<i>Scales:</i>									
Anxious/depressed	1.5 (0–10)	2 (0–19)	0.55	1 (0–7)	2 (0–9)	0.62	3 (1–10)	2 (0–19)	0.38
Withdrawn/depressed	2 (0–13)	2.5 (0–9)	0.60	1 (0–11)	3 (0–5)	0.24	7 (0–13)	2 (0–9)	0.20
Somatic complaints	1 (0–9)	0 (0–7)	0.46	1 (0–7)	0 (0–6)	0.99	1 (0–9)	0 (0–7)	0.30
Social problems	2.5 (0–10)	3 (0–14)	0.99	2 (1–10)	3 (0–6)	0.99	3 (0–6)	2 (1–14)	0.87
Thought problems	0 (0–6)	0 (0–10)	0.85	0 (0–6)	1 (0–4)	0.99	0 (0–4)	0 (0–10)	0.99
Attention problems	2.5 (0–13)	3.5 (0–15)	0.99	2 (0–9)	3 (0–8)	0.68	5 (0–13)	5 (0–15)	0.73
Rule-breaking behavior	1 (0–10)	2 (0–7)	0.36	1 (0–10)	2 (0–7)	0.34	1 (0–8)	1 (0–6)	0.79
Aggressive behavior	3 (0–13)	3 (0–16)	0.52	2 (0–13)	3 (0–13)	0.53	5 (0–9)	5 (0–16)	0.81
D. Adult self-report on emotional and behavioral problems (aged 18–41)									
ASR 18 + scales <sup>a</sup>	Study groups		p-Value	Men		p-Value	Women		p-Value
	DSD	Controls		DSD	Controls		DSD	Controls	
	n = 34	n = 34		n = 20	n = 20		n = 14	n = 14	
	Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)	
Total problems	40.5 (3–138)	32.5 (2–116)	0.40	28.5 (3–80)	25.5 (2–106)	0.53	64.5 (12–178)	53.5 (5–116)	0.24
Internalizing problems	15.5 (0–59)	10.5 (0–51)	0.04	11.5 (0–41)	7.5 (1–38)	0.15	25 (7–59)	15 (3–51)	0.15
Externalizing problems	8 (1–36)	8 (0–25)	0.65	6 (1–22)	5.5 (0–24)	0.60	11 (1–36)	14 (0–25)	0.98
<i>Scales:</i>									
Anxious/depressed	9 (0–34)	5 (0–26)	0.01	6.5 (0–23)	2.5 (0–16)	0.04	14 (0–25)	14 (5–34)	0.18
Withdrawn	4 (0–16)	2.5 (0–10)	0.07	2.5 (0–2)	2 (0–9)	0.69	6 (1–16)	3.5 (0–10)	0.02
Somatic complaints	1.5 (0–16)	2 (0–17)	0.51	1 (0–8)	1 (0–13)	0.84	3 (0–16)	3 (0–16)	0.60
Thought problems	2 (0–8)	2 (0–9)	0.54	1 (0–5)	1 (0–9)	0.73	2 (0–8)	4 (0–6)	0.81

(continued on next page)



Table 3 (continued)

D. Adult self-report on emotional and behavioral problems (aged 18–41)									
ASR 18 + scales <sup>a</sup>	Study groups		p-Value	Men		p-Value	Women		p-Value
	DSD	Controls		DSD	Controls		DSD	Controls	
	n = 34	n = 34		n = 20	n = 20		n = 14	n = 14	
	Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)		Mdn (range)	Mdn (range)	
Attention problems	4 (0–21)	5.5 (0–19)	0.33	3 (0–16)	3.5 (0–16)	0.60	7 (0–21)	6.5 (0–19)	0.40
Aggressive behavior	6 (0–21)	5 (0–15)	0.39	4 (1–16)	7.5 (0–21)	0.15	7.5 (0–21)	9 (0–15)	0.77
Rule-breaking behavior	1 (0–12)	1.5 (0–7)	0.94	1 (0–6)	1.5 (0–7)	0.45	1 (0–12)	1.5 (0–5)	0.30
Intrusive	1 (0–9)	1 (0–7)	0.87	1 (0–4)	1 (0–7)	0.76	1 (0–9)	1.5 (0–7)	0.89

Note. The paired Wilcoxon signed-rank test has been applied.

<sup>a</sup> Higher scores indicate more problems.

<sup>b</sup> One parent did not complete the CBCL; data were excluded from analysis. The child completed the YSR; these data were included.

#### Analyses in specific patient and control groups

Are there differences in emotional and behavioral problems between patients from different age groups? Table 4 shows that Internalizing problems, more specifically Anxiety/Depression and Thought Problems, were more prominent among adult patients whereas Externalizing problems were more frequently reported in children. Similar analysis in the matched control group revealed no significant differences across age groups on all scales.

Are behavioral and emotional problems more frequently reported by patients from specific diagnostic groups? Comparison of patients between diagnostic groups did not reveal any significant differences.

Do patients with DSD more often suffer from psychopathology than matched controls? In order to identify patients and controls with severe emotional and behavioral problems and psychopathology, we dichotomized the scale scores into two categories: below or above the cut off point for borderline-clinical ranges [8,9]. Statistical analyses revealed no significant differences between patient and control groups on any of the scales. This means that there were no differences between patients and controls regarding the number of individuals suffering severe psychopathology.

Do patients who never received medical treatments report more emotional and behavioral problems compared to patients who had received medical treatments? Again, analyses were performed in data split up in scale scores below and above the cut-off point. It was found that patients who had never received medical treatments reported more internalizing problems than patients who had been under clinical management (29.8% versus 19.7%;  $p = 0.047$ ).

Do patients who changed gender experienced more emotional and behavioral problems compared to patients who had not? In another study described elsewhere [14] we observed large numbers of children, adolescents and adults who had changed gender. Analyses performed for scale scores below/above the cut-off point did not reveal any differences between these patient groups, meaning that psychopathology was observed equally among patients who had changed gender and patients who had kept the gender allocated at birth.

## Discussion

This study aims to investigate emotional and behavioral problems in Indonesian patients with DSD. Due to unfamiliarity with DSD, many of these patients came under medical attention late and had lived for

Table 4

Comparison among children, adolescents, and adults with DSDs who are scored above cut points across age groups.

Scales	Children (n = 60)	Adolescents (n = 24)	Adults (n = 34)	p-Value
Total problems	11 (18.3)	3 (12.5)	7 (20.6)	0.73
Internalizing problems	9 (15.0)	4 (16.7)	16 (47.1)	<b>0.003*</b>
Externalizing problems	14 (23.3)	0	5 (14.6)	<b>0.02*</b>
<b>Scales:</b>				
Anxious/depressed	1 (1.7)	0	10 (29.4)	<b>&lt;0.001**</b>
Withdrawn/depressed	11 (18.3)	3 (12.5)	11 (32.4)	0.16
Somatic complaints	1 (1.7)	3 (12.5)	4 (11.8)	0.20
Social problems <sup>a</sup>	7 (11.7)	2 (8.3)	–	0.99
Thought problems	1 (1.7)	0	4 (11.8)	<b>0.046*</b>
Attention problems	3 (5.0)	1 (4.2)	5 (14.7)	0.24
Rule-breaking behavior	5 (8.3)	1 (4.2)	1 (2.9)	0.59
Aggressive behavior	10 (16.7)	1 (4.2)	3 (8.8)	0.26
Intrusive <sup>b</sup>	–	–	1 (2.9)	–

Note. Data presented in n (%). The Fisher Exact test was applied; significant at  $p < 0.05$ .

<sup>a</sup>  $n = 84$ . The social problem scale was only available in the CBCL and YSR.

<sup>b</sup>  $n = 34$ . The intrusive scale was only available in the ASR.

many years with an ambiguous body appearance. We hypothesized that lack of knowledge on the diagnosis, the atypical appearance of the body and reactions from the community could lead to emotional instability and would have repercussions on behavior. In this study, parents reported more emotional and behavioral problems in their children than parents of control children did. Parents reported social problems, attention problems, and, particularly in boys, aggressive behaviors and externalizing problems. Adult patients, men in particular, reported anxiety and had elevated scores on the internalizing problems scale. Women were more likely to report social isolation. They did not report anxiety, but the number of women participating in this study was smaller than the number of men and non-significant differences found in this group may be explained by insufficient sample sizes. No specific emotional and behavioral problems were reported by adolescent patients and their parents. The adolescent group was smaller than the other age groups and the small group size may explain lack of differences between adolescent patients and their control subjects. Parents of young girls with CAH indicated that their daughters tended to withdraw themselves from social activities. Although patients with DSD reported more emotional and behavioral problems than controls, severe psychopathology was not observed more frequently in the patient groups.

Parents of young boys with DSD reported more aggressive behaviors and more overall externalizing problems than parents of their matched control boys did, whereas such differences were not found among young girls. In Chinese patients with DSD, Zhu et al. [16] observed that boys with DSD displayed more depression than the control boys did. We observed higher reports of depression too, but only in adult men with DSD. These men also reported more anxiety. Compared to controls, women with DSD withdrew themselves more frequently from social activities. Living a withdrawn life is often associated with depression. Depressive complaints were also observed by Migeon et al. in American men and women with 46, XY DSD [17]. Unfortunately, we could not compare our findings on adult patients with general population of Indonesia due to lack of published data.

Emotional and behavioral problems in patients with DSD have been assessed before by Kleinemeier et al. [15]. They did not find differences between patient and normative groups. The patients participating in these studies had been under clinical management soon after identification of their DSD. Our findings in Indonesian patients therefore may be an indication that early diagnostic evaluation, information on the disorder and medical treatment may prevent the development of emotional problems.

This study is unique; for a long time patients had lived with a lack of knowledge about their condition and without sufficient medical help. Almost half of the patients (48%) never had received any information about their DSD condition nor had received medical or surgical treatments prior to the study. The remaining 52% of the patients had received some treatment, but almost all patients never received complete medical care. Often patients could not afford surgery or medication, or medication was not available regularly. All patients had to live with

ambiguous genitalia and/or an ambiguous body appearance following puberty. We assume it is difficult to cope with these ambiguities, particularly when such conditions are not understood. For some of the patients it is difficult to cope, and difficulties in coping may lead to the development of emotional problems such as depression and anxiety. The findings in this study are in line with previous findings in a study on sexuality; women with DSD reported sexual distress, fear of rejection, and avoidance to enter romantic relationships [18].

Our findings underline that it is important that patients with DSD will be referred immediately after identification of their condition to a specialized, multidisciplinary teams. These teams should not only offer diagnostic evaluation and medical treatment, but also education and counseling. These conditions are congenital and cannot be cured. In non-Western countries, for many patients, complete clinical management is not available or is too expensive. As patients have to live with DSD for a lifetime, it is important that they understand their condition and can communicate about it with their family members and/or other members of their communities. Such communication can improve understanding and acceptance and support.

In conclusion, we identified emotional instability among Indonesian patients with DSD who were identified late in life. Internalizing problems were evident among patients. Early identification of DSD and early referral to a specialized center for clinical management may contribute in understanding the condition, enhances coping abilities, and improves patients' wellbeing and therefore should be promoted among health practitioners in Indonesian primary care centers.

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#### Financial disclosure

The authors have no conflicts of interest.

#### Authors' contributions

SMHF and SLSD initiated the study and had been involved in written revisions of the manuscript. AE, AZJ, and AD designed the study and had been involved in written revisions of the manuscript. AE involved in the scale adaptation into local language and in collecting the data. AE, AD, and JE analyzed the data, produced the figures and had been involved in interpretation of findings and written the manuscript.

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#### Appendix A. 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
				Age	Phallus <sup>f</sup>	EMS <sup>g</sup>	P/Q <sup>h</sup>	Gender	
<i>Children (01–60)</i>									
01	6	46,XX DSD	CAH-SV <sup>a</sup>	3	2.8	6	3	F	None
02	6	46,XX DSD	CAH-SV <sup>a</sup>	3	4.5	4	4	F	Hydrocortisone since age 3.
03	6	46,XX DSD	CAH-SV <sup>a</sup>	6	4	4	4	F	Clitoridoplasty age 6; hydrocortisone since age 6.
04	6	46,XX DSD	CAH-SV <sup>a</sup>	4	4	7	4	F	Hydrocortisone since infancy; clitoridoplasty age 5.
05	6	46,XX DSD	CAH-SV <sup>a</sup>	6	4.5	4	4	F	Hydrocortisone since age 4 (other clinic).
06	7	46,XX DSD	CAH-SV <sup>a</sup>	2 months	2.5	4	4	F	Hydrocortisone since age 3.
07	8	46,XX DSD	CAH-SV <sup>a</sup>	4	4.1	4	4	F	Medication at birth (other clinic; details unknown); clitoridectomy age 5.
08	8	46,XX DSD	CAH-SV <sup>a</sup>	2	3	4	3	F	Hydrocortisone since age 1; clitoridectomy age 2.
09	8	46,XX DSD	CAH-SV <sup>a</sup>	8	4.3	4	4	F	None
10	8	46,XX DSD	CAH-SV <sup>a</sup>	8	6	4	5	M	Hydrocortisone irregularly since infancy.
11	8	46,XX DSD	CAH-SV <sup>a</sup>	8	5.5	3	3	M	None
12	9	46,XX DSD	CAH-SV <sup>a</sup>	7	4.5	4	4	F	None
13	10	46,XX DSD	CAH-SV <sup>a</sup>	6	4.5	4	3	F	Hydrocortisone irregularly at infancy; regularly since age 4; clitoridoplasty age 7.
14	11	46,XX DSD	CAH-SV <sup>a</sup>	7	5	4	4	F	None
15	11	46,XX DSD	CAH-SV <sup>a</sup>	11	3	7	4	F	Clitoridectomy age 2; hydrocortisone since age 11.
16	11	46,XX DSD	CAH-SV <sup>a</sup>	11	Normal clitoris	1	3	F	None
17	11	46,XX DSD	CAH-SV <sup>a</sup>	11	Normal clitoris	4	2	F	Clitoridectomy age 4 (other clinic).
18	11	46,XX DSD	CAH-SV <sup>a</sup>	11	3.5	4	3	F	Clitoridoplasty age 8 (other clinic) and 11; hydrocortisone age 11.
19	7	Chromosomal DSD	46,XX/47,XY	1	3	9	4	M	None
20	7	Chromosomal DSD	46,XX/47,XY	7	3	8.5	4	M	None
21	9	Chromosomal DSD	46,XY/46,XX	4	3.5	6	2	M	Orchidectomy age 4; HCG injection age 5

(continued on next page)



## Appendix A. (continued)

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study
				Age	Phallus <sup>f</sup>	EMS <sup>g</sup>	P/Q <sup>h</sup>	Gender	
22	6	Chromosomal DSD	46,XY/45,X <sup>c</sup>	2	1.5	6	2	M	Surgery once (details unknown)
23	7	Chromosomal DSD	46,XY/45,X	2	1.7	1	4	F	Vaginoplasty, orchidectomy age 4
24	9	Chromosomal DSD	46,XY/45,X	9	4	5	3	M	None
25	6	46,XY DSD	GD <sup>b</sup>	2	2	5	2	M	None
26	6	46,XY DSD	GD <sup>b</sup>	5	3	5	3	M	Chordaectomy age 5; urethroplasty age 6
27	7	46,XY DSD	GD <sup>b</sup>	3	3	9	3	M	Surgery age 2 (details unknown; other clinic)
28	8	46,XY DSD	GD <sup>b</sup>	7	3.5	3	3	F	None
29	10	46,XY DSD	GD <sup>b</sup>	10	Normal clitoris	1	6	F	None
30	11	46,XY DSD	GD <sup>b</sup>	10	4.5	6	2–3	M	None
31	6	46,XY DSD	PAIS <sup>c</sup>	2	2	6	2	M	Surgery (details unknown)
32	6	46,XY DSD	PAIS <sup>c</sup>	5	2.5	7	3	M	HCG injection age 5
33	10	46,XY DSD	PAIS <sup>c</sup>	4	4.9	10	2	M	None
34	10	46,XY DSD	PAIS <sup>c</sup>	10	4.5	6.5	4	M	None
35	11	46,XY DSD	PAIS <sup>c</sup>	11	4.5	6	3	M	None
36	6	46,XY DSD	Hypomasculinization <sup>d</sup>	1	2	6	3	M	Hypospadias correction twice, urethroplasty age 2
37	6	46,XY DSD	Hypomasculinization <sup>d</sup>	3	2	6	3	M	Hypospadias corrections three times (details unknown)
<b>Children (01–60)</b>									
38	6	46,XY DSD	Hypomasculinization <sup>d</sup>	4	3	5	2	M	Chordaectomy, orchidopexy age 4; urethroplasty age 5
39	6	46,XY DSD	Hypomasculinization <sup>d</sup>	6	3	6	3	M	None
40	7	46,XY DSD	Hypomasculinization <sup>d</sup>	2	3.7	5.5	4	F	None
41	8	46,XY DSD	Hypomasculinization <sup>d</sup>	7	3	4	2	M	None
42	8	46,XY DSD	Hypomasculinization <sup>d</sup>	3	3	6	2	M	Chordaectomy age 2; urethroplasty age 3 and 4 (other clinic)
43	9	46,XY DSD	Hypomasculinization <sup>d</sup>	9	3	6	3	M	None
44	10	46,XY DSD	Hypomasculinization <sup>d</sup>	7	2	6	3	M	Urethroplasty, chordaectomy, age 7 and 10
45	10	46,XY DSD	Hypomasculinization <sup>d</sup>	8	2.5	6	3	M	Surgery age 9 and 10 (details unknown)
46	11	46,XY DSD	Hypomasculinization <sup>d</sup>	7	3	6	3	M	Orchidopexy age 7
47	11	46,XY DSD	Hypomasculinization <sup>d</sup>	11	3.5	6	4	M	None
48	11	46,XY DSD	Hypomasculinization <sup>d</sup>	11	4	6	3	M	None
49	6	46,XY DSD	Hypomasculinization <sup>d</sup>	2	4	9	3	M	Surgery twice (details unknown; other clinic)
50	6	46,XY DSD	Hypomasculinization <sup>d</sup>	5	3	9	3	M	Chordaectomy, urethroplasty age 5
51	6	46,XY DSD	Hypomasculinization <sup>d</sup>	6	2.4	9	2	M	None
52	7	46,XY DSD	Hypomasculinization <sup>d</sup>	7	3	9	2	M	None
53	8	46,XY DSD	Hypomasculinization <sup>d</sup>	3	3	10	2	M	None
54	9	46,XY DSD	Hypomasculinization <sup>d</sup>	4	2.5	9	3	M	Hypospadias correction age 6
55	9	46,XY DSD	Hypomasculinization <sup>d</sup>	6	3	9	2	M	None
56	9	46,XY DSD	Hypomasculinization <sup>d</sup>	6	3	10	3	M	Surgery age 6, 7, 8 (details unknown)
57	9	46,XY DSD	Hypomasculinization <sup>d</sup>	9	(low position) 2.7	10	3	M	None
58	9	46,XY DSD	Hypomasculinization <sup>d</sup>	8	3.5	9	3	M	None
59	10	46,XY DSD	Hypomasculinization <sup>d</sup>	7	4	9	3	M	Hypospadias correction age 4, 6, 7
60	10	46,XY DSD	Hypomasculinization <sup>d</sup>	9	3.5	9	3	M	Chordaectomy age 9.
<b>Adolescents (61–84)</b>									
61	12	46,XX DSD	CAH-SV <sup>a</sup>	12	Normal clitoris	1	3	F	Clitoridectomy age 3 (other clinic).
62	16	46,XX DSD	CAH-SV <sup>a</sup>	16	Normal clitoris	1	2	F	Clitoridectomy age 7 (other clinic).
63	13	46,XX DSD	Cloacal mal-formation <sup>e</sup>	12	1.9	1	0	F	Colostomy repair one day after birth.
64	13	Chromosomal DSD	46,XY/45,X	12	3	8	2	M	Hypospadias correction, gonadectomy age 12.
65	14	Chromosomal DSD	46,XY/45,X	10	3.2	10	1	M	None
66	14	Chromosomal DSD	46,XY/45,X	13	4	8	2	M	Chordectomy age 13.
67	13	46,XY DSD	GD <sup>b</sup>	12	4.1	8	3	F	None
68	15	46,XY	GD <sup>b</sup>	15	6.5	8	2	M	None
69	17	46,XY DSD	GD <sup>b</sup>	13	Normal clitoris	1	6	F	Gonadectomy age 10 (other clinic); estrogen supplement since age 13.
70	17	46,XY DSD	GD <sup>b</sup>	17	5	4	4	F	None
71	16	46,XY DSD	CAIS <sup>c</sup>	16	Normal clitoris	1	6	F	None
72	12	46,XY DSD	PAIS <sup>c</sup>	5	1.5	7	4	M	None
73	12	46,XY DSD	PAIS <sup>c</sup>	9	3	5	3	F	None
74	13	46,XY DSD	PAIS <sup>c</sup>	12	3.1	6	3	M	None
75	15	46,XY DSD	PAIS <sup>c</sup>	12	3.1	6	5	M	Testosterone injection, gynecomastia age 13; hydrocele surgery age 14
76	12	46,XY DSD	Hypomasculinization <sup>d</sup>	12	5	6	3	M	None
77	13	46,XY DSD	Hypomasculinization <sup>d</sup>	13	5	6	3	M	Chordectomy age 5, 6
78	14	46,XY DSD	Hypomasculinization <sup>d</sup>	14	3.2	7	3	M	Surgery age 4 (details unknown); chordectomy, urethroplasty age 14
79	14	46,XY DSD	Hypomasculinization <sup>d</sup>	10	3	5	2	M	Urethroplasty age 8
80	17	46,XY DSD	Hypomasculinization <sup>d</sup>	12	7.1	6.5	3	F	None
81	17	46,XY DSD	Hypomasculinization <sup>d</sup>	14	6.5	6	2	M	Hypospadias correction age 15
82	13	46,XY DSD	Hypomasculinization <sup>d</sup>	8	4	9	2	M	Surgery (details unknown)



## Appendix A. (continued)

Patients code	Age at study	Diagnosis		Degree of masculinization at admission					Treatment received prior to study
				Age	Phallus <sup>f</sup>	EMS <sup>g</sup>	P/Q <sup>h</sup>	Gender	
83	14	46,XY DSD	Hypomasculinization <sup>d</sup>	9	5.2	9	2	M	Chordectomy, hypospadias correction age 10 and 11
84	15	46,XY DSD	Hypomasculinization <sup>d</sup>	14	5	9	2	M	Chordectomy age 14, urethroplasty age 15
<i>Adults (85–118)</i>									
85	18	46,XX DSD	CAH-SV <sup>a</sup>	11	3.6	4	3	F	Clitoridectomy age 16; Hydrocortisone
86	22	46,XX DSD	CAH-SV <sup>a</sup>	17	7	4	5	M	None
87	24	46,XX DSD	CAH-SV <sup>a</sup>	24	5	7	5	M	None
88	36	46,XX DSD	CAH-SV <sup>a</sup>	33	3.6	4	3	F	Clitoral reduction age 34; prednisone since age 36.
89	22	46,XX DSD	Cloacal malformation <sup>c</sup>	15	3.5	4	2	M	None
90	18	46,XX DSD	GD <sup>b</sup>	10	3.2	6	4	M	Treatment in other clinic (unknown), in infancy and age 7
91	18	Chromosomal DSD	46,XX/45,X	18	Normal clitoris	1	0	F	None
92	20	Chromosomal DSD	46,XX/45,X	20	Normal clitoris	1	0	F	Pills (unknown; to get menstruation) age 20; Dianne35 pills
<i>Adults (85–118)</i>									
93	29	Chromosomal DSD	45,X/46,XX	25	Normal clitoris	1	0	F	Cycloprogynova age 23; Profertil, Zumenon age 30
94	19	Chromosomal DSD	46,XY/45,X	19	5.5	6	3	F	None
95	20	Chromosomal DSD	46,XY/45,X	20	6	4	3	F	None
96	19	46,XY DSD	GD <sup>b</sup>	14	4.1	4	3	F	Gonadectomy age 14
97	19	46,XY DSD	GD <sup>b</sup>	19	Normal clitoris	2	6	F	None
98	19	46,XY DSD	GD <sup>b</sup>	19	5	8	3	F	None
99	21	46,XY DSD	GD <sup>b</sup>	14	6	5	2	F	None
100	21	46,XY DSD	GD <sup>b</sup>	21	5	4	3	F	Chordectomy age 11
101	23	46,XY DSD	GD <sup>b</sup>	23	Normal clitoris	1	7	F	None
102	27	46,XY DSD	GD <sup>b</sup>	27	5	4.5	3	F	None
103	27	46,XY DSD	GD <sup>b</sup>	26	2.5	3	4	M	None
104	39	46,XY DSD	GD <sup>b</sup>	39	1	5	5	F	None
105	41	46,XY DSD	GD <sup>b</sup>	41	3	6	4	M	None
106	24	46,XY DSD	CAIS <sup>c</sup>	24	Normal clitoris	1	6	F	None
107	18	46,XY DSD	PAIS <sup>c</sup>	11	3	5	3	M	None
108	18	46,XY DSD	PAIS <sup>c</sup>	15	6.3	7	3	M	Chordectomy age 15; urethroplasty age 16
109	20	46,XY DSD	PAIS <sup>c</sup>	16	3.7	9	2	M	Gynecomasty, chordectomy, hypospadias corrections age 14 and 15
110	27	46,XY DSD	PAIS <sup>c</sup>	27	4.5	6	4	F	None
111	31	46,XY DSD	PAIS <sup>c</sup>	26	2.6	6	3	F	Gynecomasty, chordee correction age 24; hypospadias correction age 26.
112	19	46,XY DSD	Hypomasculinization <sup>d</sup>	14	5.3	7	2	M	Chordectomy age 15; urethroplasty age 19.
113	22	46,XY DSD	Hypomasculinization <sup>d</sup>	17	1.5	3	3	F	None
114	26	46,XY DSD	Hypomasculinization <sup>d</sup>	23	5.9	5	3	F	Gynecomasty, chordectomy, hypospadias correction age 23.
115	20	46,XY DSD	Hypomasculinization <sup>d</sup>	15	6	11	2	M	Hypospadias correction age 5.
116	20	46,XY DSD	Hypomasculinization <sup>d</sup>	15	6.5	9	3	M	Penis bend (twice), hypospadias corrections age 13, 15, and 16.
117	23	46,XY DSD	Hypomasculinization <sup>d</sup>	23	6	9	2–3	M	None
118	28	46,XY DSD	Hypomasculinization <sup>d</sup>	28	6	9	2–3	M	Chordectomy age 22 (other clinic).

Note. Age in years. EMS = external masculinization score; M = male, F = female.

<sup>a</sup> 46,XX CAH-SV = congenital adrenal hyperplasia simple virilization type. CYP 21 mutation was confirmed in all patients [4].

<sup>b</sup> 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.

<sup>c</sup> 46 XY DSD CAIS/PAIS = complete/partial androgen insensitivity syndromes. A mutation in the AR gene was confirmed [4].

<sup>d</sup> 46 XY DSD hypomasculinization refers to hypomasculinization with unknown cause. Molecular and biochemical details are described in other publication elsewhere [4]

<sup>e</sup> Cloacal exstrophy.

<sup>f</sup> in centimeters (cm).

<sup>g</sup> Degree of masculinization based on external genital features, ranged from 0 to 12.

<sup>h</sup> P/Q = Prader and Quigley stages; stands for degree of genital masculinization in 46, XX (Prader) and 46, XY (Quigley) individuals [19,20].

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