



## 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	Mosaics	3	0	3	6
	46, XY DSD	5	5	6	16
	Gonadal dysgenesis <sup>b</sup>	9	7	12	28
46, XX DSD	Hypomasculinization e.c.i.	25	9	7	41
	CAH – SV <sup>c</sup>	18	2	4	24
	Gonadal dysgenesis	0	0	1	1
	Cloacal malformation	0	1	1	2
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

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