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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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ABSTRACT

It has been assumed that physical atypicality will enhance vulnerability for social stigmatization in patients with disorders or differences of sex development (DSD). In Indonesia, until recently diagnostic evaluation and treatment was sparsely available. We assessed social stigmatization in Indonesian patients who had lived in body ambiguity.

Eighty-one parents of youngsters with DSD (aged 6-17 years), and 34 adult patients with DSD (aged 18-41 years) filled out the Social Stigmatization Scale towards DSD (SSS-DSD). The majority of patients have not received any hormonal or surgical interventions prior to the study. Differences in reported stigmatization were explored across gender, gender change history, treatment status, and visibility of DSD characteristics.

Patients with atypical genitals, easily identifiable atypical physical appearance, patients who displayed cross gender behaviour and patients who changed gender experienced social stigmatization. Rejection elicited depression in females and children and adolescents who had changed gender. In text analysis five themes were identified that explained stigmatization and stress: ignorance on DSD, patient's personality, patient's responses, cultural norms and society's response towards patients with DSD.

Particularly patients who had not been able to conceal their condition (patients with physical atypicality and patients who changed gender) experienced social stigmatization. Stigmatization was stressful and related to isolation. Education on DSD, self-empowerment and medical interventions to prevent atypical physical appearance may remove barriers for acceptance.

KEYWORDS: social stigmatization, DSD, intersexuality, gender atypicality, body atypicality, Indonesia

INTRODUCTION

Disorders or differences of sex development (DSD) refer to a group of congenital conditions in which development of chromosomal, gonadal, or anatomical sex is atypical, often leading to an atypical appearance of the genitals and body (1). Clinicians specialised in DSD are confronted with parents' and patients' difficulties to cope the atypical appearance and the derogatory reactions their physical atypicality may elicit. In addition to treatments necessary for survival, clinical management aims to reduce or prevent physical atypicality and to enable sexual functioning in order to increase the patient's opportunities for social participation. These interventions are criticized, as they have great impact on the child's life and are taken without involvement of the child him-/herself. It has been argued that such interventions do not give room to variety in sex and gender and are principally conducted to comfort parents or support the gender ideology of society (2-6). Calls have been made to stop this practice (7-9). Yet, there is a lack of systematic data on DSD-associated stigma (2). Randomized controlled comparison between early gender assignment, genital corrections and hormonal interventions and delayed interventions is highly valued (10) but is difficult to conduct. Despite criticism, most parents living in Western countries choose gender assignment and genital surgery for their children with DSD (11,12). Follow-up studies on quality of life are scarce and findings are inconsistent (13-15). Medical literature only contains a few reports on DSD and social stigmatization (16-24).

In Indonesia, DSD is not widely known among health practitioners and laymen. Clinical management had been challenged by limited diagnostic and treatment facilities. Many patients had to live with atypical bodies and had been raised with doubts about their gender (25,26). This enables us to investigate their experiences of being raised in ambiguity (25,26) and of social stigmatization.

METHODS

Study design and setting

Experiences with social stigmatization due to DSD were evaluated from adult patients and parents of children and adolescents. Data collection was carried out between March 2007 and May 2011. All patients consulted the DSD Team of the Dr. Kariadi Hospital. The study protocol was approved by the board of the ethical committee at the Faculty of Medicine, Diponegoro University, Semarang, Indonesia.

Patients

All patients with a proven diagnosis of DSD consulting the DSD Team of the Dr. Kariadi Hospital (27) were invited for study participation. Patients and parents received oral and written study information (provided by AZJ) and had given their informed consent. Patients with a genital anomaly and additional features suggestive of a dysmorphic syndrome (28), patients with sex chromosome DSD without mosaicism, and patients with DSD and intellectual disabilities (indicated from the child's academic achievements and/or observed by the medical doctor in interaction with the patient) were excluded. Thirty-four adults (20 men; 14 women; aged 18-41 years) and 81 parents of 60 children (42 boys, 18 girls; aged 6-11) and 21 adolescents (15 boys; 6 girls; aged 12-17 years) took part in the study. Table 1 summarizes patient characteristics and diagnoses.

Table 1. DSD diagnoses of participants in the study (N=115)

DSD diagnosis		Age			
		6-11	12-17	18+	Total
Sex chromosome DSD	Patients with 45X/46XY; 46XidicY; 46,XX/46,XY; 46,XX/47,XXY	6	3	5	14
46 XY DSD	AIS*	5	5	6	16
	Gonadal dysgenesis †	6	2	10	18
	Hypomasculinization¥	25	9	7	41
46 XX DSD	CAH – SV‡	18	2	4	24
	Gonadal dysgenesis †	-	-	1	1
	Cloacal malformation	-	-	1	1
Total		60	21	34	115

* Androgen Insensitivity syndrome. AR gene mutation was confirmed (27).

† Abnormal hormonal testicular function with uni/bilaterally undescended testes. The clinical and biochemical presentation suggest gonadal dysfunction. Serum levels of luteinizing hormone and follicle stimulating hormone were elevated but testosterone, anti-müllerian hormone and Inhibin are low for age, and no or diminished serum testosterone response to HCG.

¥ 46 XY karyotype with hypomasculinization of unknown cause, despite extensive analysis (27)

‡ Simple virilising type of congenital adrenal hyperplasia. CYP 21 mutation was confirmed (27).

Details on diagnosis and degree of masculinization at admission per patient can be found in Ediati. et al. (14,25)

Procedure

After parents and adult patients had given their written consent, psychological assessment including data on patient's socio-economic and ethnic-cultural background (14,25,26). was conducted in the hospital or at the patient's home, by a trained psychologist (AE).

Materials

Prior to this study, no measure was available to assess social stigmatization in patients with DSD. Therefore, we developed the Social Stigmatization Scale for DSD (SSS-DSD). The SSS-DSD assesses the frequency of experienced stigmatization and the level of stress-level evoked by the stigmatizing experiences using a Likert scale with response mode ranging from '*not at all*' (1) to '*very much*' (5). In addition, we asked patients to explain their

experiences and their beliefs on the cause of DSD, their worries, and ability to cope with DSD. We developed parental and adult versions of the SSS-DSD.

The applicability of the SSS-DSD was tested (by AE) prior to implementation and revealed that applying the measure as a paper-pencil test was feasible for well-educated subjects. For parents and patients with low educational levels the measure preferably was applied orally.

Data analysis

Construct validity of both the adult and parental versions of the SSS-DSD scale was explored using principal component analysis (PCA) with varimax rotation and Kaiser Normalization method. Factors with Eigen values greater than 1 and items with factor loadings (after rotation) greater than 0.40 were considered acceptable. Instrument reliability was evaluated as internal consistency with Cronbach's Alpha as outcome measure.

The overall and domain sum scores of the SSS-DSD were calculated as the unweight sum scores of the individual domains and items, respectively. With Spearman's correlation coefficient (*rho*) the correlations between different types of experienced stigma and evoked stress were evaluated. The Kruskal-Wallis test was applied to test for differences in continuous data of more than two groups, the Mann-Whitney U test for differences between two independent groups. Differences in categorical data were compared using Fisher's Exact test. Differences were considered significant at $p < .05$ (two-sided).

RESULTS

The majority of participants was male, lived in rural areas, was Javanese and Moslem, parents' educational background varied from no formal education to university level, the

majority had attended high school and worked in the lower-income sector or were unemployed. Details on socio-economic and ethnic-cultural variables can be found in Table 2.

Table 2. Participant characteristics (N=115)

Characteristics	Children and adolescents (*n*=81)	Adults (*n*=34)
Gender (of patients)		
Male	57 (70·4)	20 (58·8)
Female	24 (29·6)	14 (41·2)
Treatment		
Received treatment [a]	44 (54·3)	15 (44·1)
No treatment	37 (45·7)	19 (55·9)
Social gender role change		
Yes	7 (8·6)	15 (44·1)
No	74 (91·4)	19 (55·9)
Visibility of DSD [b]		
Visible	12 (14·8)	17 (50·0)
Partly hidden	57 (70·4)	17 (50·0)
Hidden	12 (14·8)	
Region		
Central Java	70 (86·4)	29 (85·2)
Other provinces in Java	8 (9·9)	2 (5·9)
Outside Java island	3 (3·7)	3 (8·8)
Ethnic		
Javanese	76 (93·8)	31 (91·2)
Non Javanese	5 (6·2)	3 (8·8)
Religion		
Islam	77 (95·1)	33 (97·1)
Non Islam	4 (4·9)	1 (2·9)
Residential setting		
Rural	45 (55·6)	15 (44·1)
Suburban	24 (29·6)	11 (32·4)
Urban	12 (14·8)	8 (23·5)
Highest education attained	(Fathers* / Mothers*)	(Adults)
No formal education	9 (11·3) / 10 (12·5)	4 (11·8)
Elementary school	27 (33·7) / 28 (35·0)	3 (8·8)
High school	36 (45·0) / 36 (45·0)	23 (67·6)
University	8 (10·0) / 6 (7·5)	4 (11·8)
Parents' occupation	(Fathers* / Mothers*)	(Adults)
Unemployed	0 / 44 (55·0)	13 (38·2)
Labour	47 (58·7) / 22 (27·5)	9 (26·5)
Self employed	16 (20·0) / 6 (7·5)	4 (11·8)
Staff	17 (21·3) / 8 (10·0)	8 (23·5)

Data are presented in *n* (%) * One father/mother missing for being deceased.

[a] Treatment in most patients had been minimal, for instance, patients had taken glucocorticoid therapy for only a limited period or had undergone one surgical procedure for hypospadias correction when two or more procedures were needed (14, 25-27)

[b] Visibility of DSD refer to all those aspects of physical and behavioral atypicality that cannot be hidden in social interaction. Concealable refer to physical atypicality that can be covered by clothes (partly hidden) and non-ambiguous phenotype (hidden).

Validity and reliability of SSS-DSD parent and adult versions

SSS-DSD Parent. The PCA extracted four components explaining 56% of the total variance: a) stigmatization elicited by genital ambiguity (items 1-2, 5-6, 11; α = 0·86); b) stigmatization elicited by ambiguous body appearance or gender role behaviour (items 3-4, 7-8a; α = 0·84); c) social rejection (items 9-10, 12; α = 0·88); and d) emotional problems due to DSD (items 13a-d, 13g-h; α = 0·85). Table 3a shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity and reliability of the SS-DSD Parent were considered satisfactory.

Table 3*a.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Parental report (*n*=81)

Questions	Components			
	1[a]	2[b]	3[c]	4[d]
01a. Can other people see that your child got a genital that is (slightly) different from that of other children?	**0·60**	0·44	0·07	-0·14
01b. How stressful is this to you?	**0·72**	0·22	0·31	-0·09
02a. Do you think that other people look at your child because of the atypical genital?	**0·64**	0·38	-0·05	0·19
02b. How stressful is this to you?	**0·73**	0·13	0·17	0·21
05a. Do other people speak negatively about *your child* because of the atypical genital or physical appearance?	**0·65**	-0·05	0·27	0·24
05b. How stressful is this to you?	**0·67**	-0·10	0·34	0·14
06a. Do people speak negatively about *you* because of your child?	**0·76**	-0·07	-0·18	0·13
06b. How stressful is this to you?	**0·73**	-0·11	-0·17	0·15
11a. Is your child called names or teased by other children because of child's atypical genital or physical appearance?	**0·40**	0·03	0·24	0·55
11b. How stressful is this to you?	**0·41**	0·02	0·44	0·49
03a. Can other people see that your child has an atypical physical appearance?	-0·09	**0·76**	0·07	0·35
03b. How stressful is this to you?	-0·14	**0·52**	0·18	0·48
04a. Do you think that other people look at your child because of the atypical physical appearance?	0·39	**0·67**	-0·21	0·16
04b. How stressful is this to you?	0·17	**0·57**	0·13	-0·01
07a. Does your child show more cross gender role behaviour compared to other children? For parents of daughters: Does your daughter prefer more masculine activities than other girls? For parents of sons: Does your son prefer more feminine activities compared to other boys?	-0·06	**0·87**	0·10	0·20
07b. How stressful is this to you?	-0·01	**0·91**	0·04	0·02
08a. Do other people speak or behave negatively about your child because of child's cross gender role behavior behaviour? (Daughters: masculine behaviour and interests? Sons: feminine behaviour and interests?)	0·11	**0·44**	-0·08	-0·05

09a. Do other people isolate or reject *your child* because of atypical genital/physical appearance?	-0·03	0·34	**0·76**	0·19
09b. How stressful is this to you?	0·04	0·24	**0·85**	0·03
10a. Do other people isolate or reject *you* because of your child?	0·17	-0·10	**0·86**	-0·13
10b. How stressful is this to you?	0·21	-0·12	**0·82**	-0·14
12a. Is your child isolated or rejected by other children because of the atypical genital or physical appearance?	-0·09	0·00	**0·75**	0·45
12b. How stressful is this to you?	0·02	-0·08	**0·88**	0·22
13a. Does your child suffer from emotional problems because of the atypical genital or physical appearance?	0·26	0·00	-0·07	**0·75**
13b. How stressful is this to you?	0·13	0·02	-0·03	**0·82**
13c. How frequent was your child sad?	0·09	0·06	-0·05	**0·55**
13d. How frequent was your child depressed?	0·01	-0·01	0·07	**0·82**
13g. How frequent was your child shy?	-0·14	0·14	0·13	**0·71**
13h. How frequent was your child socially withdrawn?	-0·11	0·34	0·13	**0·61**
13e. How frequent was your child angry?	0·01	0·19	0·20	0·37
13f. How frequent was your child aggressive?	0·12	0·05	0·28	0·24
14. Are you worried about your child's future?	0·20	-0·01	0·05	0·29
15. Is it difficult for you to accept your child?	0·25	0·07	0·02	-0·12

[a] Stigmatization due to genital ambiguity and stress evoked by such experiences (α = 0·86).
[b] Stigmatization due to atypical physical appearance or displayed cross-gender role behaviour and stress evoked by such experiences (α = 0·84).
[c] Social rejection or isolation due to DSD and stress evoked by being rejected or isolated (α = 0·88).
[d] Reported emotional problems seen in the child and parental stress evoked these emotional problems (α = 0·85).

SSS-DSD Adult. The PCA extracted three components explaining 62.9% of the total variance: a) verbal stigmatization (items 1-2, 4-5, 7; α = 0·92); b) behavioural stigmatization (items 3, 6a, 9-10; α = 0·85); and c) emotional problems due to DSD (items 13-15; α = 0·94). Table 3b shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity and reliability of the SSS-DSD Adult were also considered satisfactory.

Table 3*b.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Adult report (*n*=34)

Questions	Components		
	Verbal [a]	Behaviour [b]	Emotion [c]
1a. Can other people see that you got a genital that is (slightly) different from that of other men/women?	**0·63**	0·03	0·10
1b. How stressful is this to you?	**0·62**	0·36	0·42
2a. Do you think that other people look at you because of the atypical genital?	**0·79**	0·33	0·22
2b. How stressful is this to you?	**0·86**	0·19	0·23
4a. Do you think that other people look at you because of the atypical appearance?	**0·71**	-0·08	0·37
4b. How stressful is this to you?	**0·82**	0·21	0·25

5a. Do other people speak negatively about you because of the atypical genital or physical appearance?	**0·75**	0·08	-0·13
5b. How stressful is this to you?	**0·86**	0·10	-0·05
7a. Do other people including family member speak or behave negatively about you because of you show more cross-gender behaviour compared to others? (For woman: Do you prefer more masculine activities compared to other women? For man: do you prefer more feminine activities compared to other men?)	**0·73**	-0·27	0·12
7b. How stressful is this to you?	**0·71**	-0·23	0·12
3a. Can other people see that you have an atypical appearance?	0·08	**0·65**	0·41
3b. How stressful is this to you?	0·43	**0·55**	0·23
6a. Do you behave differently from other men/women?	0·01	**0·64**	0·17
6b. How stressful is this to you?	0·16	0·36	0·10
9a. Do other people teased you or called you by funny names because of the atypical genital or physical appearance?	0·10	**0·84**	0·07
9b. How stressful is this to you?	0·15	**0·87**	0·18
10a. Do other people isolate/reject you because of the atypical genital or physical appearance?	-0·21	**0·68**	0·23
10b. How stressful is this to you?	-0·21	**0·68**	0·23
13a. Do you suffer from emotional problems because of the atypical genital/appearance?	0·31	0·40	**0·75**
13b. How stressful is this to you?	0·31	0·37	**0·75**
13c. How frequently you were sad?	-0·06	0·20	**0·94**
13d. How frequently you were depressed?	0·11	0·16	**0·93**
13e. How frequently you were angry?	0·34	0·31	**0·68**
13g. How frequently you were shy?	0·17	0·16	**0·73**
13h. How frequently you were socially withdrawn?	0·11	0·14	**0·71**
14. Are you worried about your future?	0·21	0·10	**0·74**
15. Is it difficult for you to accept your condition?	-0·02	0·20	**0·75**

[a] Verbal reaction received due to DSD conditions and the stress evoked by such experiences (α = 0·92).
[b] Behavioural reaction received due to DSD conditions and the stress evoked by such experiences (α = 0·85).
[c] Reported emotional problem due to having DSD conditions (α = 0·94).

Correlations between stigmatization and stress

In both measures, items measuring experiences with stigmatization were positively and significantly correlated with items measuring stress evoked by such stigmatization, in all components measured.

SSS-DSD Parent. Stigmatization due to genital ambiguity positively correlated with stress (r_s (79) = 0·794, $p < 0·001$); stigmatization elicited by an ambiguous appearance or behaviour positively correlated with stress (r_s (79) = 0·80, $p < 0·001$); social rejection positively correlated with stress (r_s (79) = 0·81, $p < 0·001$); and emotional problems also positively correlated with stress (r_s (79) = 0·64, $p < 0·001$).

SSS-DSD Adult. Verbal stigmatization positively correlated with stress (r_s (32) = 0·755, $p < 0·001$); behavioural stigmatization positively correlated with stress (r_s (32) = 0·753, $p < 0·001$); and emotional and acceptance problems due to DSD also positively correlated with stress (r_s (32) = 0·882, $p < 0·001$). The more frequent patients experienced social stigmatization, the higher the stress.

Subgroup analysis

Tables 4a and 4b summarize the comparisons across gender, treatment status, gender change history, and visibility of DSD conditions. In either boys or girls, children and adolescents experienced some degree of stigmatization. Girls reported more stigmatization due to ambiguous appearance or cross-gender role behaviour and had more emotional problems than boys (see Table 4a; gender comparison). Women experienced more stigmatization and had more emotional problems than men. Both men and women experienced some degree of verbal and behavioural reactions due to their DSD conditions (see Table 4b; gender comparison).

Regardless of having received prior hormonal/surgical treatment for DSD, children and adolescents experienced stigmatization and had emotional problems (see Table 4a; treatment status comparison). However, untreated adults experienced more stigmatization than treated adults (see Table 4b; treatment status comparison).

Six youngsters and 15 adults were assigned female at birth but had changed gender later in life (25). These patients experienced more stigmatization than patients who kept their initial gender. Young people and adults experienced more stigmatization due to ambiguous appearance, cross gender role behaviour or behavioural stigmatization and had more emotional problems than youngsters who kept the initial gender (see Table 4a / 4b; gender

change comparison). Adults experienced more behavioural stigmatization than adults who kept the gender assigned at birth (see Table 4b; gender change history comparison).

Children and adolescents with visible ambiguity of the body experienced stigmatizations more frequently than patients who could conceal ambiguous characteristics (see Table 4a; visibility of DSD comparison). Regardless of the visibility of DSD, children and adolescents reported emotional problems due to DSD. Adults with visible ambiguity of the body experienced more stigmatization than adults who could conceal ambiguity; this was particularly seen in verbal and behavioural stigmatization (see Table 4b; visibility of DSD comparison).

Table *4a.* Median domain and overall sum scores of the SS-DSD Parent-Report across gender, treatment status, gender change history, and visibility of DSD*

SSS-DSD Parent-report	**Gender**			**Treatment status**			**Gender change history**			**Visibility of DSD**		
	Boys[a] (*n* = 57)	Girls (*n* = 24)	*p*	Treated (*n* = 54)	Untreated (*n* = 27)	*p*	Yes[b] (*n* = 6)	No (*n* = 75)	*p*	Visible (*n* = 12)	Concealable[c] (*n* = 69)	*p*
Atypical genital	12 (10-50)	10 (10-27)	0·48	11 (10-50)	15 (10-37)	0·20	16 (10-23)	12 (10-50)	0·26	18 (10-32)	11 (10-50)	**0·006**
Atypical appearance/behaviour	10 (10-14)	10 (10-30)	**<0·001**	10 (10-23)	10 (10-30)	0·42	11 (10-30)	10 (10-23)	**0·01**	14 (10-30)	10 (10-21)	**<0·001**
Social rejection	10 (10-37)	10 (10-23)	0·26	10 (10-37)	10 (10)	0·14	10 (10)	10 (10-37)	0·99	10 (10-37)	10 (10-22)	**0·01**
Emotional problems	10 (10-28)	10 (10-32)	**0·002**	10 (10-32)	10 (10-22)	0·44	14 (10-22)	10 (10-32)	**0·02**	10 (10-32)	10 (10-32)	0·11
Total score[d]	43 (40-103)	49 (40-98)	0·23	43 (40-103)	45 (40-74)	0·47	54 (45-74)	42 (40-103)	**0·02**	61 (40-98)	42 (40-103)	**0·002**

* * Data were presented as median (range). The Mann-Whitney U test was applied.

[a a] The terms men and women are accordingly to the gender the patient presented himself socially and to us when he or she participated in the study.

[b b] Ediati A. et al. (25)

[c c] Visible refer to all those aspects of physical and behavioural atypicality that cannot be hidden in social interaction. Concealable refer to physical atypicality that can be covered by clothes (partly hidden) and typical phenotype (hidden).

[d d] Unweight sum score.

Table 4b Median domain and overall sum scores of the SSS-DSD Adult-Report across gender, treatment status, gender change history, and visibility of DSD*

SSS-DSD Adult-report	Gender			Treatment status			Gender change history			Visibility of DSD		
	Men[a] (n = 20)	Women (n = 14)	p	Treated (n = 15)	Untreated (n = 19)	p	Yes[b] (n = 15)	No (n = 19)	p	Visible (n = 17)	Concealable[c] (n = 17)	p
Verbal stigmatization	12 (10-47)	10 (10-28)	0·78	10 (10-36)	14 (10-47)	0·14	14 (10-47)	10 (10-28)	0·11	15 (10-47)	10 (10-21)	**0·015**
Behavioural stigmatization	10 (10-26)	11 (10-37)	0·30	10 (10-23)	10 (10-37)	0·25	11 (10-31)	10 (10-37)	**0·03**	13 (10-37)	10 (10-11)	**0·001**
Emotional problems	14 (10-40)	29 (10-47)	**0·009**	12 (10-40)	21 (10-47)	0·07	16 (10-47)	10 (10-44)	0·52	16 (10-47)	16 (10-40)	0·60
Total score[d]	37 (30-91)	50 (31-100)	**0.042**	36 (30-83)	48 (31-100)	**0·046**	41 (31-100)	38 (30-97)	0·80	46 (31-100)	38 (30-63)	0·19

* Data were presented as median (range). The Mann-Whitney U test was applied.

[a] The terms men and women are accordingly to the gender the patient presented himself socially and to us when he or she participated in the study,

[b] Ediati A. et al. (15)

[c] Visible refer to all those aspects of physical and behavioural atypicality that cannot be hidden in social interaction . Concealable refer to physical ambiguity that can be covered by clothes (partly hidden) and atypical phenotype (hidden).

[d] Unweight sum score

DISCUSSION

Our study revealed that atypical appearance of the genitals and / or body is problematic and hard to cope with. Stigmatization was most prominent in patients with an ambiguous appearance who could not hide their ambiguity, in untreated adult patients, in patients who changed their gender, and in females. The more frequently they experienced stigmatization due to DSD, the higher their stress. Patients who were able to hide features of body atypicality from others did not report less emotional problems than patients who had visible features of DSD. This suggests that fear and prevention of being stigmatised is as problematic as having experienced stigmatization. From the qualitative data we found out that a substantial number of patients withdrew themselves from social interactions, such as withdrawal from school and avoiding interaction with neighbours or community members. In Indonesia a hostile attitude towards those who show variant sex or gender development is often met; patients are humiliated and excluded.

Overall, many patients did not give high rates for experienced social stigmatization. This may indicate that these patients did not experience social stigmatization. It is also possible that patients gave answers they considered appropriate. Our findings show that especially, patients with an atypical appearance of the genitals or the body due to DSD deal with incomprehension and social stigmatization. The social stigmatization is stressful and affects their psychosocial wellbeing. The study supports the assumption that an atypical appearance can be adverse for social participation and quality of life. Does the study also support medical interventions that will prevent development or progression of an ambiguous appearance, such as removal of (underdeveloped) gonads, and hormone replacement therapy in patients with a 46, XY DSD? This question is more complicated and difficult to answer. Part of the social stigmatization was related to lack of knowledge on DSD among patients themselves and Indonesian layman. We assume that stigmatization can be prevented or reduced by education. Self-empowerment will amplify patients' and parents' ability to cope with DSD. Educated patient and parents can educate their social network and help them to

improve their position in the community (29). In addition, educated patients and parents will be better able to decide which treatments are optimal for their particular circumstances.

Indonesia is a collective society in which procreation and progeny are highly valued. Some people with DSD cannot meet such expectations (14,25,26). Our findings are in line with previous studies reporting sexual distress, disclosure dilemmas, and tendency to avoid romantic relationships among women with DSD (26). Women with DSD report a more vulnerable position than affected men in this culture. This may explain why we recruited more male patients (59%) than female patients (41%) for this study. This study includes 20 patients who underwent a female-to-male gender change, 16 of them initiated a gender change in adolescence or adulthood. Three patients had a 46 XX, karyotype, 17 patients had a 46, XY karyotype (25). Progressive masculinization may have induced gender dysphoria and instigated the wish to change gender, but ostracism may also contribute to gender change for people.

Limited assessment of the construct validity of the SSS-DSD was considered as the study limitation. Our study focussed on the relationship between social stigmatization and atypical consequently to delay of treatment. As no suitable measure was available, we developed one. In developing a measure, it is preferred to perform cross-validation studies next to principal component analysis to assess construct validity more extensively. Unfortunately, quantitative measures to assess different aspects of psychosocial wellbeing, were unavailable in Indonesia and we were unable to perform such analyses (14,25,26).

This study is relevant for patients with DSD who face delay of treatment due to poor understanding of their medical condition, inadequate laboratory support and lack of appropriate and affordable medications (22). Although social stigmatization emerges in the interaction between the individual and society and Asian societies differ from Western societies, the study results are also relevant for Western patients. Although Western culture is individually centered and demands to follow social norms (e.g. giving birth) are less

stringent, Western patients with DSD, due to their sexual ambiguity, have a vulnerable position in society too. We aim to optimize patients' psychosexual and psychosocial wellbeing and are searching for adaptations in clinical management (13).

CONCLUSION

Patients with DSD, and particularly those with an atypical sexual appearance, are prone to stigmatization. Such stigmatization is stressful and leads to emotional reactions. These findings support the assumption that an atypical appearance of the body is harmful for psychosocial wellbeing. Medical interventions to prevent progressive development of an atypical appearance may facilitate patient's self-esteem and social participation. In addition, education about DSD that is culturally sensitive and accessible for patients and laymen, may remove barriers for acceptance.

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Authors' Contributions: SMHF and SD initiated the study. SMHF, SD and AW had been involved in written revisions of the manuscript, AE, AD and EB designed the study, analysed the data, produced the figures and performed literature searches and written revisions. AE, JO and AD developed the questionnaires, AE collected the data, AdlC assisted in qualitative data analysis, had been involved in interpretation of qualitative data and written revisions

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1 Box

What this known about the subject

- DSD is a somatically and socially challenging condition; many patients and parents suffer from emotional problems, experience or anticipate social stigmatization
- Opposing opinions rule the debate on how to strengthen patients' emotional wellbeing and improve their psychosocial opportunities
- At present DSD-associated social stigma has not been investigated systematically. Such studies are necessary in order to make proper adjustments in clinical management

What this paper adds

- Upon patients' and parents' individual reports of ostracism in Indonesia, we developed the Social Stigmatization Scale for DSD and investigated patients' and parents' experienced stigma
- Experienced and anticipated DSD related stigmatization was highest among patients with body atypicality and patients who changed gender
- Social stigmatization was evaluated as stressful, related to (self)isolation and highly correlated with depression.

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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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ABSTRACT

Objectives: To access social stigmatization related to atypical appearance of the body, including but not limited to the external genitalia, among Indonesian patients with a disorder of sex development (DSD). Until recently, diagnostic evaluation, information about the underlying causes of DSD and treatment options were sparsely available for these patients.

Methods: Eighty-one parents of children and adolescents with DSD (aged 6-17 years), and 34 adult patients with DSD (aged 18-41 years) completed the Social Stigmatization Scale towards DSD (SSS-DSD), an instrument developed to assesses the frequency of stigmatization and the level of stress associated with these experiences. Open-ended questions investigated detailed information on stigmatization as well as parents' and patients' emotional and behavioural reactions to these experiences. Differences in stigmatization were explored across sex of rearing, gender change history, treatment status, and DSD characteristics that could be easily identified by others (e.g. masculinisation of the body in females).

Results: Social stigmatization was reported by patients with atypical appearance of their genitalia, atypical appearance of their body aside from their genitals, among those who displayed cross-gender behaviour and those who changed gender. Among participants reared female, and among children and adolescents who changed gender, social stigmatization was associated with ostracism, depressive symptoms and social isolation.

Conclusions: Patients unable to conceal their condition (those with visible physical atypicality and those who changed gender) experienced social stigmatization. Stigmatization was stressful and related to isolation and withdrawal from social interaction. Education about DSD, self-empowerment and medical interventions to prevent atypical physical development may remove barriers to acceptance by others for affected individuals.

KEYWORDS: social stigmatization, DSD, intersexuality, gender atypicality, body atypicality, Indonesia

INTRODUCTION

Disorders/ of sex development (DSD) refer to a group of congenital conditions in which development of chromosomal, gonadal, or anatomical sex is atypical, often leading to an atypical appearance of the genitals and other parts of the body that differ in appearance between males and females(1). Clinicians specialised in DSD treatment are confronted with parents' and patients' difficulties in coping with the atypical physical development and the derogatory reactions their atypicality may elicit. In addition to treatments necessary for survival, clinical management aims to reduce or prevent physical atypicality and to enable sexual functioning in order to increase the patient's opportunities for social participation. These interventions have been criticized, as they impact the child's life and are often performed without the child's assent or consent. It has been argued that such interventions do not allow for diversity in sex and gender development and are principally conducted to comfort parents or support the gender ideology of society (2-6). As such, calls have been made by some to stop this practice of medical and surgical intervention (7-9); however, there is a lack of systematic data on DSD-associated stigma among affected individuals who did not receive such interventions (2). Randomized, controlled studies of early gender assignment, genital surgery and hormonal interventions compared to delayed interventions is highly valued (10) but difficult to conduct. Despite criticisms noted above, most parents living in Western countries choose early gender assignment and surgical correction of the atypical genitalia for their children with DSD (11,12). Follow-up studies on quality of life are scarce and findings are inconsistent regarding the risks and benefits of medical intervention (13-15). Finally, the medical literature contains few reports on DSD and social stigmatisation (16-23).

In Indonesia, DSD is not widely known among health practitioners and laymen. Clinical management is challenged by limited diagnostic and treatment facilities. As a result,

many patients live with atypical bodies and experience doubts about their gender (24,25). During outpatient clinic visits, experiences with social stigmatisation were often reported spontaneously by these patients and stimulated many patients and parents to seek medical help. This enabled us to investigate these patients' experiences of living with physical ambiguity and doubts about their gender (24,25), as well as their experience of social stigmatisation.

METHODS

Study design and setting

Experiences with social stigmatization due to DSD were evaluated from adult patients and parents of affected children and adolescents. Data collection was carried out between March 2007 and May 2011. All patients consulted the DSD Team of the Dr. Kariadi Hospital. The study protocol was approved by the board of the ethical committee at the Faculty of Medicine, Diponegoro University, Semarang, Indonesia.

Patients

All patients with a confirmed diagnosis of DSD consulting the DSD Team of the Dr. Kariadi Hospital (26) were invited for study participation. Patients and parents received oral and written study information (provided by AZJ) and provided informed consent. Patients with a genital anomaly and additional features suggestive of a dysmorphic syndrome (27), patients with sex chromosome DSD without mosaicism, and patients with DSD and intellectual disabilities (indicated from the child's academic achievements and/or observed by the medical doctor in interaction with the patient) were excluded. Thirty-four adults (20 men; 14 women; aged 18-41 years) and 81 parents of 60 children (42 boys, 18 girls; aged 6-11) and

21 adolescents (15 boys; 6 girls; aged 12-17 years) participated, with a participation rate of 78%. Table 1 summarizes patient characteristics and diagnoses.

Table 1. DSD diagnoses of participants in the study (N=115)

DSD diagnosis		Age			
		6-11	12-17	18+	Total
Sex chromosome DSD	Patients with 45X/46XY; 46XidicY; 46,XX/46,XY; 46,XX/47,XXY	6	3	5	14
46 XY DSD	AIS*	5	5	6	16
	Gonadal dysgenesis †	6	2	10	18
	Hypomasculinization¥	25	9	7	41
46 XX DSD	CAH – SV‡	18	2	4	24
	Gonadal dysgenesis †	-	-	1	1
	Cloacal malformation	-	-	1	1
Total		60	21	34	115

* Androgen Insensitivity syndrome. AR gene mutation was confirmed (27).
† Abnormal hormonal testicular function with uni/bilaterally undescended testes. The clinical and biochemical presentation suggest gonadal dysfunction. Serum levels of luteinizing hormone and follicle stimulating hormone were elevated but testosterone, anti-müllerian hormone and Inhibin are low for age, and no or diminished serum testosterone response to HCG.
¥ 46 XY karyotype with hypomasculinization of unknown cause, despite extensive analysis (27)
‡ Simple virilising type of congenital adrenal hyperplasia. CYP 21 mutation was confirmed (27).
Details on diagnosis and degree of masculinization at admission per patient can be found in Ediati. et al. (14,25)

Procedure

After obtaining written, informed consent, psychological assessment including data on patients' socio-economic and ethnic-cultural background (14,24,25) was collected in the hospital or at the patient's home, by a trained psychologist (AE).

Materials

Prior to this study, no measure was available to assess social stigmatization in patients with DSD. Therefore, we developed the Social Stigmatization Scale for DSD (SSS-DSD). The SSS-DSD assesses the frequency of experienced stigmatization (1-13a. questions) and

the level of stress evoked by the stigmatizing experiences (1-13b. questions) using a Likert scale with responses ranging from '*not at all*' (1) to '*very much*' (5). In addition, we asked patients to give details about their experiences with DSD, their beliefs on the cause of their DSD, their concerns and ability to cope with DSD (1-12c. questions). We developed parent and adult versions of the SSS-DSD.

The applicability of the SSS-DSD was tested (by AE) prior to implementation and revealed that applying the measure as a paper-pencil test was feasible for well-educated subjects. The rating scale was piloted in a small group of 20 patients and parents with DSD. After a few adaptations, the SSS-DSD seemed suitable for application in this study. Formal large scale psychometric pretesting among sizable numbers of patients or their parents was considered unfeasible in view of the limited numbers of patients with rare genetic conditions. For parents and patients with low educational levels the measure preferably was applied verbally.

Data analysis

Construct validity of both the adult and parental versions of the SSS-DSD scale was explored using principal component analysis (PCA) with varimax rotation and Kaiser Normalization method. Factors with eigenvalues greater than 1 and items with factor loadings (after rotation) greater than 0.40 were considered acceptable. Instrument reliability was evaluated as internal consistency with Cronbach's Alpha as outcome measure.

The overall and domain sum scores of the SSS-DSD were calculated as the unweighted sum scores of the individual domains and items, respectively. For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion emotional problems. With Spearman's correlation coefficient (*rho*) the correlations between different types of stigma and evoked stress were evaluated. The Kruskal-Wallis test was

applied to test for differences in continuous data of more than two groups, the Mann-Whitney U test for differences between two independent groups. Differences in categorical data were compared using Fisher's Exact test. Differences were considered significant at $p < .05$ (two-sided).

Qualitative data collected were analysed by inductive content analyses using NVivo qualitative data analysis software (28,29). AE started an open coding procedure and finally clustered codes into 4 themes. Relationships between themes were investigated using the compound coding application in NVivo (28,29).

RESULTS

The majority of participants were male, lived in rural areas, Javanese and Muslim. Parents' educational backgrounds varied from no formal education to university level, and the majority attended high school and worked in the lower-income sector or were unemployed. Details on socio-economic and ethnic-cultural variables can be found in Table 2.

Table 2. Participant characteristics (N=115)

Characteristics	Children and adolescents (*n*=81)	Adults (*n*=34)
Gender (of patients)		
Male	57 (70·4)	20 (58·8)
Female	24 (29·6)	14 (41·2)
Treatment		
Received treatment [a]	44 (54·3)	15 (44·1)
No treatment	37 (45·7)	19 (55·9)
Social gender role change		
Yes	7 (8·6)	15 (44·1)
No	74 (91·4)	19 (55·9)
Visibility of DSD [b]		
Visible	12 (14·8)	17 (50·0)
Partly hidden	57 (70·4)	17 (50·0)
Hidden	12 (14·8)	
Region		
Central Java	70 (86·4)	29 (85·2)
Other provinces in Java	8 (9·9)	2 (5·9)
Outside Java island	3 (3·7)	3 (8·8)
Ethnic		
Javanese	76 (93·8)	31 (91·2)
Non Javanese	5 (6·2)	3 (8·8)
Religion		
Islam	77 (95·1)	33 (97·1)
Non Islam	4 (4·9)	1 (2·9)
Residential setting		
Rural	45 (55·6)	15 (44·1)
Suburban	24 (29·6)	11 (32·4)
Urban	12 (14·8)	8 (23·5)
Highest education attained	(Fathers* / Mothers*)	(Adults)
No formal education	9 (11·3) / 10 (12·5)	4 (11·8)
Elementary school	27 (33·7) / 28 (35·0)	3 (8·8)
High school	36 (45·0) / 36 (45·0)	23 (67·6)
University	8 (10·0) / 6 (7·5)	4 (11·8)
Parents' occupation	(Fathers* / Mothers*)	(Adults)
Unemployed	0 / 44 (55·0)	13 (38·2)
Labour	47 (58·7) / 22 (27·5)	9 (26·5)
Self employed	16 (20·0) / 6 (7·5)	4 (11·8)
Staff	17 (21·3) / 8 (10·0)	8 (23·5)

Data are presented in *n* (%) * One father/mother missing for being deceased.

[a] Treatment in most patients had been minimal, for instance, patients had taken glucocorticoid therapy for only a limited period or had undergone one surgical procedure for hypospadias correction when two or more procedures were recommended (14, 25-27)

[b] Social gender role change could be physician imposed, parent imposed or patient initiated (25)

[c] Visibility of DSD refer to all those aspects of physical and behavioural atypicality that cannot be hidden during social interaction. Concealable refers to physical atypicality that can be covered by clothes (partly hidden) and non-ambiguous phenotype (hidden).

Reliability and validity of SSS-DSD parent and adult versions

SSS-DSD Parent. The PCA extracted four components with Cronbach's alphas ranging between 0.84-0.88. Reliability (internal consistency) of the Parent version can be considered as good. The four components explaining 56% of the total variance were as following: a) stigmatization elicited by genital ambiguity (items 1-2, 5-6, 11; α = 0·86); b) stigmatization elicited by atypical physical appearance or cross-gender role behaviour (items 3-4, 7-8a; α = 0·84); c) social exclusion (items 9-10, 12; α = 0·88); and d) emotional problems due to DSD (items 13a-d, 13g-h; α = 0·85). Table 3a shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity of the SS-DSD Parent were considered satisfactory.

Table 3*a.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Parental report (*n*=81)

Questions	Components			
	1[a]	2[b]	3[c]	4[d]
Stigma elicited by genital atypicality				
01a. Can other people see that your child has genitals that are (slightly) different from that of other children?	**0·60**	0·44	0·07	-0·14
01b. How stressful is this to you?	**0·72**	0·22	0·31	-0·09
01c[e]. Open-ended question: Can you tell us more about this?				
02a. Do you think that other people look at your child because of their atypical genitalia?	**0·64**	0·38	-0·05	0·19
02b. How stressful is this to you?	**0·73**	0·13	0·17	0·21
05a. Do other people speak negatively about *your child* because of their atypical genital or physical appearance?	**0·65**	-0·05	0·27	0·24
05b. How stressful is this to you?	**0·67**	-0·10	0·34	0·14
06a. Do people speak negatively about *you* because of your child?	**0·76**	-0·07	-0·18	0·13
06b. How stressful is this to you?	**0·73**	-0·11	-0·17	0·15
11a. Is your child called names or teased by other children because of their atypical genital or physical appearance?	**0·40**	0·03	0·24	0·55
11b. How stressful is this to you?	**0·41**	0·02	0·44	0·49
Stigma elicited by physical atypicality or cross gender role behaviour				
03a. Can other people see that your child has an atypical physical appearance?	-0·09	**0·76**	0·07	0·35
03b. How stressful is this to you?	-0·14	**0·52**	0·18	0·48
04a. Do you think that other people look at your child because of their atypical physical appearance?	0·39	**0·67**	-0·21	0·16
04b. How stressful is this to you?	0·17	**0·57**	0·13	-0·01
07a. Does your child show more cross-gender role behaviour compared to other children? For parents of daughters: Does your daughter prefer more masculine activities than other girls? For parents of sons: Does your son prefer more feminine activities compared to other boys?	-0·06	**0·87**	0·10	0·20
07b. How stressful is this to you?	-0·01	**0·91**	0·04	0·02

08a. Do other people speak or behave negatively about your child because of their cross-gender role behaviour? (Daughters: masculine behaviour and interests? Sons: feminine behaviour and interests?)	0·11	**0·44**	-0·08	-0·05
Experiences with social exclusion				
09a. Do other people isolate *your child* because of atypical of their genital/physical appearance?	-0·03	0·34	**0·76**	0·19
09b. How stressful is this to you?	0·04	0·24	**0·85**	0·03
10a. Do other people isolate *you* because of your child?	0·17	-0·10	**0·86**	-0·13
10b. How stressful is this to you?	0·21	-0·12	**0·82**	-0·14
12a. Is your child isolated by other children because of their atypical genital or physical appearance?	-0·09	0·00	**0·75**	0·45
12b. How stressful is this to you?	0·02	-0·08	**0·88**	0·22
Emotional reactions				
13a. Does your child suffer from emotional problems because of their atypical genital or physical appearance?	0·26	0·00	-0·07	**0·75**
13b. How stressful is this to you?	0·13	0·02	-0·03	**0·82**
13c. How frequent was your child sad?	0·09	0·06	-0·05	**0·55**
13d. How frequent was your child depressed?	0·01	-0·01	0·07	**0·82**
13g. How frequent was your child shy?	-0·14	0·14	0·13	**0·71**
13h. How frequent was your child socially withdrawn?	-0·11	0·34	0·13	**0·61**
13e. How frequent was your child angry?	0·01	0·19	0·20	0·37
13f. How frequent was your child aggressive?	0·12	0·05	0·28	0·24
14. Are you worried about your child's future?	0·20	-0·01	0·05	0·29
15. Is it difficult for you to accept your child?	0·25	0·07	0·02	-0·12

[a] Stigmatization due to genital ambiguity and stress evoked by such experiences (α = 0·86).
[b] Stigmatization due to atypical physical appearance or displayed cross-gender role behaviour and stress evoked by such experiences (α = 0·84).
[c] Social exclusion or isolation due to DSD and stress evoked by being rejected or isolated (α = 0·88).
[d] Reported emotional problems seen in the child and parental stress evoked these emotional problems (α = 0·85).
[e] Each question 1-12 was followed by an open-ended question: Can you tell us more about this? Example?

SSS-DSD Adult. The PCA extracted three components with Cronbach's alphas ranging between 0.85-0.94. Reliability (internal consistency) of the Adult version can be considered as good to very good. The extracted three components explaining 62.9% of the total variance were the following: a) verbal stigmatization (items 1-2, 4-5, 7; α = 0·92); b) behavioural stigmatization (items 3, 6a, 9-10; α = 0·85); and c) emotional problems due to DSD (items 13-15; α = 0·94). Table 3b shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity of the SSS-DSD Adult were also considered satisfactory.

Table 3*b.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Adult report (*n*=34)

Questions	Components		
	Verbal [a]	Behaviour [b]	Emotion [c]
Verbal stigmatisation			
1a. Can other people see that you have genitalia that are (slightly) different from other men/women?	**0·63**	0·03	0·10
1b. How stressful is this to you?	**0·62**	0·36	0·42
1c[d]. Open-ended question: Can you tell us more about this?			
2a. Do you think that other people look at you because of your atypical genitalia?	**0·79**	0·33	0·22
2b. How stressful is this to you?	**0·86**	0·19	0·23
4a. Do you think that other people look at you because of your atypical physical appearance?	**0·71**	-0·08	0·37
4b. How stressful is this to you?	**0·82**	0·21	0·25
5a. Do other people speak negatively about you because of your atypical genital or physical appearance?	**0·75**	0·08	-0·13
5b. How stressful is this to you?	**0·86**	0·10	-0·05
7a. Do other people, including family members, speak or behave negatively about you because you show more cross-gender behaviour compared to others? (For woman: Do you prefer more masculine activities compared to other women? For man: do you prefer more feminine activities compared to other men?)	**0·73**	-0·27	0·12
7b. How stressful is this to you?	**0·71**	-0·23	0·12
Behavioural stigmatisation			
3a. Can other people see that you have an atypical appearance?	0·08	**0·65**	0·41
3b. How stressful is this to you?	0·43	**0·55**	0·23
6a. Do you behave (slightly) differently from other men/women?	0·01	**0·64**	0·17
6b. How stressful is this to you?	0·16	0·36	0·10
9a. Do other people tease you or call you by funny names because of your atypical genital or physical appearance?	0·10	**0·84**	0·07
9b. How stressful is this to you?	0·15	**0·87**	0·18
10a. Do other people isolate/reject you because of your atypical genital or physical appearance?	-0·21	**0·68**	0·23
10b. How stressful is this to you?	-0·21	**0·68**	0·23
Emotional problems			
13a. Do you suffer from emotional problems because of your atypical genital/appearance?	0·31	0·40	**0·75**
13b. How stressful is this to you?	0·31	0·37	**0·75**
13c. How frequently were you sad?	-0·06	0·20	**0·94**
13d. How frequently were you depressed?	0·11	0·16	**0·93**
13e. How frequently were you angry?	0·34	0·31	**0·68**
13g. How frequently were you shy?	0·17	0·16	**0·73**
13h. How frequently were you socially withdrawn?	0·11	0·14	**0·71**
14. Are you worried about your future?	0·21	0·10	**0·74**
15. Is it difficult for you to accept your condition?	-0·02	0·20	**0·75**

[a] Verbal reaction received due to DSD conditions and the stress evoked by such experiences (α = 0·92).
[b] Behavioural reaction received due to DSD conditions and the stress evoked by such experiences (α = 0·85).
[c] Reported emotional problem due to having DSD conditions (α = 0·94).
[d] Each question 1-7,9,10 was followed by an open-ended question: Can you tell us more about this? Example?

Correlations between stigmatization and stress

In both measures, items measuring experiences with stigmatisation were positively and significantly correlated with items measuring stress evoked by such stigmatisation, in all components measured.

SSS-DSD Parent. Stigmatisation due to genital ambiguity positively correlated with stress (r_s (79) = 0·794, $p < 0·001$); stigmatisation elicited by an ambiguous appearance or behaviour positively correlated with stress (r_s (79) = 0·80, $p < 0·001$); social rejection positively correlated with stress (r_s (79) = 0·81, $p < 0·001$); and emotional problems also positively correlated with stress (r_s (79) = 0·64, $p < 0·001$).

SSS-DSD Adult. Verbal stigmatisation positively correlated with stress (r_s (32) = 0·755, $p < 0·001$); behavioural stigmatisation positively correlated with stress (r_s (32) = 0·753, $p < 0·001$); and emotional and acceptance problems due to DSD also positively correlated with stress (r_s (32) = 0·882, $p < 0·001$). The more frequent patients experienced social stigmatisation, the higher their reported stress.

Subgroup analysis

Tables 4a and 4b summarize the comparisons across sex of rearing, treatment status, history gender change (24), and visibility of DSD conditions. In both boys and girls, children and adolescents experienced some degree of stigmatization. Girls reported more stigmatization due to atypical physical appearance or cross-gender role behaviour and had more emotional problems than boys (see Table 4a; gender comparison). Women experienced more stigmatization and had more emotional problems than men. Both men and women experienced some degree of verbal and behavioural reactions due to their DSD conditions (see Table 4b; gender comparison).

Regardless of having received prior hormonal/surgical treatment for DSD, children and adolescents experienced stigmatization and had emotional problems (see Table 4a; treatment status comparison). However, untreated adults experienced more stigmatization than treated adults (see Table 4b; treatment status comparison).

Six youngsters and 15 adults were assigned female at birth but changed gender later in life (24). These patients experienced more stigmatization than patients who kept their initial gender. Young people and adults experienced more stigmatization due to an ambiguous appearance or cross-gender behaviour and had more emotional problems than youngsters who kept the initial sex of rearing (see Table 4a / 4b; social gender role change comparison). Adults who changed gender experienced more behavioural stigmatization than adults who kept the gender assigned at birth (see Table 4b; gender change history comparison).

Children and adolescents with visible ambiguity of the body experienced stigmatization more frequently than patients who could conceal ambiguous characteristics (see Table 4a; visibility of DSD comparison). Regardless of the visibility of DSD, children and adolescents reported emotional problems due to DSD. Adults with visible ambiguity of the body experienced more stigmatization than adults who could conceal ambiguity; this was particularly seen in verbal and behavioural stigmatization (see Table 4b; visibility of DSD comparison).

Table *4a.* Median domain and overall sum scores of the SS-DSD Parent-Report across sex of rearing, treatment status, gender change history, and visibility of DSD

SSS-DSD Parent-report	**Sex of rearing**			**Treatment status**			**Social gender role change history**			**Visibility of DSD**		
	Boys [a] (*n* = 57) median (range)	Girls (*n* = 24) median (range)	*P* [b]	Treated (*n* = 54) median (range)	Untreated (*n* = 27) median (range)	*p*	Yes [c] (*n* = 6) median (range)	No (*n* = 75) median (range)	*p*	Visible (*n* = 12) median (range)	Concealable [d] (*n* = 69) median (range)	*p*
Atypical genitalia score range 10–50	12 [e] (10-50)	10 (10-27)	0·48	11 (10-50)	15 (10-37)	0·20	16 (10-23)	12 (10-50)	0·26	18 (10-32)	11 (10-50)	**0·006**
Atypical appearance/behaviour score range 7-35	10 (10-14)	10 (10-30)	**<0·001**	10 (10-23)	10 (10-30)	0·42	11 (10-30)	10 (10-23)	**0·01**	14 (10-30)	10 (10-21)	**<0·001**
Social exclusion score range 6-30	10 (10-37)	10 (10-23)	0·26	10 (10-37)	10 (10)	0·14	10 (10)	10 (10-37)	0·99	10 (10-37)	10 (10-22)	**0·01**
Emotional problems score range10-50	10 (10-28)	10 (10-32)	**0·002**	10 (10-32)	10 (10-22)	0·44	14 (10-22)	10 (10-32)	**0·02**	10 (10-32)	10 (10-32)	0·11
Total score [f] score range 33-165	43 (40-103)	49 (40-98)	0·23	43 (40-103)	45 (40-74)	0·47	54 (45-74)	42 (40-103)	**0·02**	61 (40-98)	42 (40-103)	**0·002**

[a] The terms men and women are used according to the gender the patient presented himself or herself socially and to us when he or she participated in the study.
[b] The Mann-Whitney U test was applied.
[c] Ediati A. et al. (24)
[d] Visible refer to all those aspects of physical and behavioural atypicality that cannot be hidden in social interaction. Concealable refer to physical atypicality that can be covered by clothes (partly hidden) and typical phenotype (hidden).
[e] For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion emotional problems.
[f] Unweight sum score.

Table 4b Median domain and overall sum scores of the SSS-DSD Adult-Report across sex of rearing, treatment status, gender change history, and visibility of DSD

SSS-DSD Adult-report	**Sex of rearing**			**Treatment status**			**Social gender role change history**			**Visibility of DSD**		
	Men [a] (n = 20) median (range)	Women (n = 14) median (range)	p [b]	Treated (n = 15) median (range)	Untreated (n = 19) median (range)	p	Yes [c] (n = 15) median (range)	No (n = 19) median (range)	p	Visible (n = 17) median (range)	Concealable [d] (n = 17) median (range))	p
Verbal stigmatization [e] score range 10-50	12 (10-47)	10 (10-28)	0·78	10 (10-36)	14 (10-47)	0·14	14 (10-47)	10 (10-28)	0·11	15 (10-47)	10 (10-21)	**0·015**
Behavioural stigmatization score range 8-40	10 (10-26)	11 (10-37)	0·30	10 (10-23)	10 (10-37)	0·25	11 (10-31)	10 (10-37)	**0·03**	13 (10-37)	10 (10-11)	**0·001**
Emotional problems score range 9-45	14 (10-40)	29 (10-47)	**0·009**	12 (10-40)	21 (10-47)	0·07	16 (10-47)	10 (10-44)	0·52	16 (10-47)	16 (10-40)	0·60
Total score [f] score range 17-135	37 (30-91)	50 (31-100)	**0.042**	36 (30-83)	48 (31-10)	**0·046**	41 (31-100	38 (30-97)	0·80	46 (31-100)	38 (30-63)	0·19

[a] The terms men and women are used according to the gender the patient presented himself socially and to us when he or she participated in the study
[b] The Mann-Whitney U test was applied.
[c] Ediati A. et al. (15)
[d] Visible refer to all those aspects of physical and behavioural atypicality that cannot be hidden in social interaction . Concealable refer to physical ambiguity that can be covered by clothes (partly hidden) and atypical phenotype (hidden).
[e] For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion emotional problems.
[f] Unweight sum score

Qualitative data

In text analyses 4 themes were identified that gave insight in characteristics of social stigmatization and related stress: *a.* (correct, incorrect or lack of) knowledge about DSD, *b.* patients' personality and related emotional and behavioural responses, *c.* cultural norms and related social expectancies and *d.* response from the community.

DISCUSSION

Our study revealed that atypical appearance of the genitals and / or body is problematic (14). Stigmatization was most prominent in patients with an atypical physical appearance who could not hide their ambiguity, in untreated adult patients, in patients who changed their social gender, and in females. The more frequently they experienced DSD-related social stigma, the higher their stress. Patients who were able to hide features of body atypicality from others did not report less emotional problems than patients who had visible features of DSD. This suggests that fear and prevention of being stigmatised is as problematic as having experienced stigmatization. From the qualitative data we observed that a substantial number of patients withdrew themselves from social interactions, such as withdrawal from school and avoiding interaction with neighbours or community members. In Indonesia a hostile attitude towards those who show variant sex or gender development is often met; patients are humiliated and excluded.

Overall, many patients did not give high rates for experienced social stigmatizatiom; however, patients with atypical physical appearance are vulnerable for social stigmatization. They indicated that stigmatization was stressful, elicited negative emotions, hampered social participation and hence affected overall psychosocial wellbeing. Part of their social stigmatization was related to lack of knowledge about DSD among patients themselves and among Indonesian layman. We propose that stigmatization can be prevented or reduced by education. Similar to many other non-western countries, Indonesia has few well-trained

medical psychologists available for counselling to help patients and parents cope with DSD. Once educated and supported, patients and parents can then educate their social network to improve their position in the community (30). In addition, educated patients and parents will be better able to decide which treatments are optimal for their particular circumstances.

Indonesia is a collective society in which procreation and progeny are highly valued. Some people with DSD cannot meet such expectations (14,24,25). Our findings are in line with previous studies reporting sexual distress, disclosure dilemmas, and tendency to avoid romantic relationships among women with DSD (25). Women with DSD report a more vulnerable position than affected men in this culture. This may explain why we recruited more male patients (59%) than female patients (41%) for this study. This study includes 20 patients who underwent a female-to-male social gender change, 4 patients changed gender in childhood, 16 of them initiated a change in adolescence or adulthood. Three patients had a 46, XX karyotype and CAH, 17 patients had a 46, XY karyotype (24).

Progressive masculinization may have induced gender dysphoria and instigated the wish to change gender, but ostracism may also contribute to change for people.

Limited assessment of the construct validity of the SSS-DSD is a study limitation. Our study focussed on the relationship between social stigmatization and atypical appearance resulting from the delay of medical and surgical treatment. As no suitable measure was available, we developed one. In developing a measure, it is preferred to perform cross-validation studies in addition to principal component analysis to assess construct validity more extensively. Unfortunately, quantitative measures to assess different aspects of psychosocial wellbeing were unavailable in Indonesia and we were unable to perform such analyses (14,24,25).

This study is relevant for patients with DSD who face delay of treatment due to poor understanding of their medical condition, inadequate laboratory support and lack of appropriate and affordable medications (21). Although Western culture is individual-centered

and the demands to follow social norms (e.g. giving birth for females) are less stringent, Western patients with DSD have a vulnerable position in society too. Thus, the current results may be informative to patients and families outside of Indonesia. Ultimately, we aim to optimize patients' psychosexual and psychosocial wellbeing and are searching for adaptations in clinical management that are evidence-based, such as the reduction of stigmatization of those affected by DSD (13).

CONCLUSION

Patients with DSD, particularly those with an atypical appearance, are prone to stigmatization. Such stigmatization is stressful and leads to negative emotional reactions and social isolation. These findings support the assumption that an atypical physical appearance can be harmful for psychosocial wellbeing. This may be particularly true when the medical condition is not understood by the patient, the parents and members of the community, as well as when the patient cannot make their own decisions regarding clinical management. We assume that culturally sensitive education about DSD that is accessible for patients and laymen may remove barriers for social acceptance.

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Authors' Contributions: SMHF and SD initiated the study. SMHF, SD and AW had been involved in written revisions of the manuscript, AE, AD and EB designed the study, analysed the data, produced the figures and performed literature searches and written revisions. AE, JO and AD developed the questionnaires, AE collected the data.

Availability of data and materials

We do not wish to share data originating from our database in order to protect the anonymity of subjects included in this survey. Permission has not been obtained to share data widely with other investigators and would require individual content/assent.

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Box

What this known about the subject

- DSD is a somatically and socially challenging condition; many patients and parents suffer from emotional problems, experience or anticipate social stigmatization
- Opposing opinions rule the debate on how to strengthen patients' emotional wellbeing and improve their psychosocial opportunities
- At present DSD-associated social stigma has not been investigated systematically. Such studies are necessary in order to make proper adjustments in clinical management

What this paper adds

- We developed the Social Stigmatization Scale for DSD and investigated patients' and parents' experienced stigma
- Experienced and anticipated DSD related stigmatization was highest among patients with body atypicality and patients who changed gender
- Social stigmatization was evaluated as stressful, related to (self)isolation and highly correlated with depression.

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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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ABSTRACT

Objectives: To assess social stigmatization related to atypical appearance of the body, including, but not limited to the external genitalia, among Indonesian patients with a disorder of sex development (DSD). Until recently, diagnostic evaluation, information about the underlying causes of DSD and treatment options were sparsely available for these patients.

Methods: Eighty-one parents of children and adolescents with DSD (aged 6-17 years), and 34 adult patients with DSD (aged 18-41 years) completed the Social Stigmatization Scale towards DSD (SSS-DSD), an instrument developed to assesses the frequency of stigmatization and the level of stress associated with these experiences. Open-ended questions investigated detailed information on stigmatization as well as parents' and patients' emotional and behavioural reactions to these experiences. Differences in stigmatization were explored across sex of rearing, gender change history, treatment status, and DSD characteristics that could be easily identified by others (e.g. masculinisation of the body in females).

Results: Social stigmatization was reported by patients with atypical appearance of their genitalia, atypical appearance of their body aside from their genitals, among those who displayed cross-gender behaviour and those who changed gender. Among participants reared as female, and among children and adolescents who changed gender, social stigmatization was associated with ostracism, depressive symptoms and social isolation.

Conclusions: Patients unable to conceal their condition (those with visible physical atypicality and those who changed gender) experienced social stigmatization. Stigmatization was stressful and related to isolation and withdrawal from social interaction. Education about DSD, self-empowerment and medical interventions to prevent atypical physical development may remove barriers to acceptance by others for affected individuals.

KEYWORDS: social stigmatization, DSD, intersexuality, gender atypicality, body atypicality, Indonesia

INTRODUCTION

Disorders of sex development (DSD) refer to a group of congenital conditions in which development of chromosomal, gonadal, or anatomical sex is atypical, often leading to an atypical appearance of the genitals and other parts of the body that differ in appearance between males and females(1). Clinicians specialised in DSD treatment are confronted with parents' and patients' difficulties in coping with the atypical physical development and the derogatory reactions their atypicality may elicit. In addition to treatments necessary for survival, clinical management aims to reduce or prevent physical atypicality and to enable sexual functioning in order to increase the patient's opportunities for social participation. These interventions have been criticized, as they impact the child's life and are often performed without the child's assent or consent. It has been argued that such interventions do not allow for diversity in sex and gender development and are principally conducted to comfort parents or support the gender ideology of society (2-6). As such, there have been calls to stop this practice of medical and surgical intervention (7-9). However, there is a lack of systematic data on DSD-associated stigma among affected individuals who did not receive such interventions (2). Randomized, controlled studies of early gender assignment, genital surgery and hormonal interventions compared to delayed interventions are highly valued (10) but difficult to conduct. Despite criticisms noted above, most parents living in Western countries choose early gender assignment and surgical correction of the atypical genitalia for their children with DSD (11,12). Follow-up studies on quality of life are scarce and findings are inconsistent regarding the risks and benefits of medical intervention (13-15). Finally, the medical literature contains few reports on DSD and social stigmatisation (16-23).

In Indonesia, DSD is not widely known among health practitioners and laymen. Clinical management is challenged by limited diagnostic and treatment facilities. As a result, many patients live with atypical bodies and experience doubts about their gender (24,25).

During outpatient clinic visits, experiences with social stigmatisation were often reported spontaneously by these patients and stimulated many patients and parents to seek medical help. This enabled us to investigate these patients' experiences of living with physical ambiguity and doubts about their gender (24,25), as well as their experience of social stigmatisation.

METHODS

Study design and setting

Experiences with social stigmatization due to DSD were evaluated from adult patients and parents of affected children and adolescents. Data collection was carried out between March 2007 and May 2011. All patients consulted the DSD Team of the Dr. Kariadi Hospital. The study protocol was approved by the board of the ethical committee at the Faculty of Medicine, Diponegoro University, Semarang, Indonesia.

Patients

All patients with a confirmed diagnosis of DSD consulting the DSD Team of the Dr. Kariadi Hospital (26) were invited to participate in the study. Patients and parents received oral and written study information (provided by AZJ) and provided informed consent. Patients with a genital anomaly and additional features suggestive of a dysmorphic syndrome (27), patients with sex chromosome DSD without mosaicism, and patients with DSD and intellectual disabilities (indicated from the child's academic achievements and/or observed by the medical doctor in interaction with the patient) were excluded. Thirty-four adults (20 men; 14 women; aged 18-41 years) and 81 parents of 60 children (42 boys, 18 girls; aged 6-11) and

21 adolescents (15 boys; 6 girls; aged 12-17 years) participated, with a participation rate of 78%. Table 1 summarizes patient characteristics and diagnoses.

Table 1. DSD diagnoses of participants in the study (N=115)

DSD diagnosis		Age			
		6-11	12-17	18+	Total
Sex chromosome DSD	Patients with 45X/46XY; 46XidicY; 46,XX/46,XY; 46,XX/47,XXY	6	3	5	14
46 XY DSD	AIS*	5	5	6	16
	Gonadal dysgenesis †	6	2	10	18
	Hypomasculinization¥	25	9	7	41
46 XX DSD	CAH – SV‡	18	2	4	24
	Gonadal dysgenesis †	-	-	1	1
	Cloacal malformation	-	-	1	1
Total		60	21	34	115

* Androgen Insensitivity syndrome. AR gene mutation was confirmed (26).
† Abnormal hormonal testicular function with uni/bilaterally undescended testes. The clinical and biochemical presentation suggest gonadal dysfunction. Serum levels of luteinizing hormone and follicle stimulating hormone were elevated but testosterone, anti-müllerian hormone and Inhibin are low for age, and no or diminished serum testosterone response to HCG.
¥ 46 XY karyotype with hypomasculinization of unknown cause, despite extensive analysis (26)
‡ Simple virilising type of congenital adrenal hyperplasia. CYP 21 mutation was confirmed (26).
Details on diagnosis and degree of masculinization at admission per patient can be found in Ediati. et al. (14,24)

Procedure

After obtaining written, informed consent, psychological assessment including data on patients' socio-economic and ethnic-cultural background (14,24,25) was collected in the hospital or at the patient's home, by a trained psychologist (AE).

Materials

Prior to this study, no measure was available to assess social stigmatization in patients with DSD. Therefore, we developed the Social Stigmatization Scale for DSD (SSS-DSD). The SSS-DSD assesses the frequency of experienced stigmatization (1-13a. questions) and

the level of stress evoked by the stigmatizing experiences (1-13b. questions) using a Likert scale with responses ranging from '*not at all*' (1) to '*very much*' (5). In addition, we asked patients to give details about their experiences with DSD, their beliefs on the cause of their DSD, their concerns and ability to cope with DSD (1-12c. questions). We developed parent and adult versions of the SSS-DSD.

The applicability of the SSS-DSD was tested (by AE) prior to implementation and revealed that applying the measure as a paper-pencil test was feasible for well-educated subjects. The rating scale was piloted in a small group of 20 patients and parents with DSD. After a few adaptations, the SSS-DSD seemed suitable for application in this study. Formal large scale psychometric pretesting among sizable numbers of patients or their parents was considered unfeasible in view of the limited numbers of patients with rare genetic conditions. For parents and patients who could not read well, the measure was applied verbally.

Data analysis

Construct validity of both the adult and parental versions of the SSS-DSD scale was explored using principal component analysis (PCA) with varimax rotation and Kaiser Normalization method. Factors with eigenvalues greater than 1 and items with factor loadings (after rotation) greater than 0.40 were considered acceptable. The reliability of the instrument was evaluated using Cronbach's Alpha as a measure of internal consistency.

The overall and domain sum scores of the SSS-DSD were calculated as the unweighted sum scores of the individual domains and items, respectively. For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion and emotional problems. With Spearman's correlation coefficient (*rho*) the correlations between different types of stigma and evoked stress were evaluated. The Kruskal-Wallis test was applied to test for differences in continuous data of more than two groups, the Mann-Whitney

U test for differences between two independent groups. Differences in categorical data were compared using Fisher's Exact test. Differences were considered significant at $p < .05$ (two-sided).

Qualitative data collected were analysed by inductive content analyses using NVivo qualitative data analysis software (28,29). AE started an open coding procedure and finally clustered codes into 4 themes. Relationships between themes were investigated using the compound coding application in NVivo (28,29).

RESULTS

The majority of participants were male, lived in rural areas, Javanese and Muslim. Parents' educational backgrounds varied from no formal education to university level, and the majority attended high school and worked in the lower-income sector or were unemployed. Details on socio-economic and ethnic-cultural variables can be found in Table 2.

Table 2. Participant characteristics (N=115)

Characteristics	Children and adolescents (*n*=81)	Adults (*n*=34)
Gender (of patients)		
Male	57 (70.4)	20 (58.8)
Female	24 (29.6)	14 (41.2)
Treatment		
Received treatment [a]	44 (54.3)	15 (44.1)
No treatment	37 (45.7)	19 (55.9)
Social gender role change		
Yes	7 (8.6)	15 (44.1)
No	74 (91.4)	19 (55.9)
Visibility of DSD [b]		
Visible	12 (14.8)	17 (50.0)
Partly hidden	57 (70.4)	17 (50.0)
Hidden	12 (14.8)	
Region		
Central Java	70 (86.4)	29 (85.2)
Other provinces in Java	8 (9.9)	2 (5.9)
Outside Java island	3 (3.7)	3 (8.8)
Ethnic		
Javanese	76 (93.8)	31 (91.2)
Non Javanese	5 (6.2)	3 (8.8)
Religion		
Islam	77 (95.1)	33 (97.1)
Non Islam	4 (4.9)	1 (2.9)
Residential setting		
Rural	45 (55.6)	15 (44.1)
Suburban	24 (29.6)	11 (32.4)
Urban	12 (14.8)	8 (23.5)
Highest education attained	(Fathers* / Mothers*)	(Adults)
No formal education	9 (11.3) / 10 (12.5)	4 (11.8)
Elementary school	27 (33.7) / 28 (35.0)	3 (8.8)
High school	36 (45.0) / 36 (45.0)	23 (67.6)
University	8 (10.0) / 6 (7.5)	4 (11.8)
Parents' occupation	(Fathers* / Mothers*)	(Adults)
Unemployed	0 / 44 (55.0)	13 (38.2)
Labour	47 (58.7) / 22 (27.5)	9 (26.5)
Self employed	16 (20.0) / 6 (7.5)	4 (11.8)
Staff	17 (21.3) / 8 (10.0)	8 (23.5)

Data are presented in *n* (%) * One father/mother missing for being deceased.

[a] Treatment in most patients had been minimal, for instance, patients had taken glucocorticoid therapy for only a limited period or had undergone one surgical procedure for hypospadias correction when two or more procedures were recommended (14, 25-27)

[b] Social gender role change could be physician imposed, parent imposed or patient initiated (25)

[c] Visibility of DSD refer to all those aspects of physical and behavioural atypicality that cannot be hidden during social interaction. Concealable refers to physical atypicality that can be covered by clothes (partly hidden) and non-ambiguous phenotype (hidden).

Reliability and validity of SSS-DSD parent and adult versions

SSS-DSD Parent. The PCA extracted four components with Cronbach's alphas ranging between 0.84-0.88. Reliability (internal consistency) of the Parent version can be considered as good. The four components explaining 56% of the total variance were as follows: a) stigmatization elicited by genital ambiguity (items 1-2, 5-6, 11; α = 0.86); b) stigmatization elicited by atypical physical appearance or cross-gender role behaviour (items 3-4, 7-8a; α = 0.84); c) social exclusion (items 9-10, 12; α = 0.88); and d) emotional problems due to DSD (items 13a-d, 13g-h; α = 0.85). Table 3a shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity of the SS-DSD Parent was considered satisfactory.

Table 3*a.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Parental report (*n*=81)

Questions	Components			
	1[a]	2[b]	3[c]	4[d]
Stigma elicited by genital atypicality [a]				
01a. Can other people see that your child has genitals that are (slightly) different from that of other children?	**0.60**	0.44	0.07	-0.14
01b. How stressful is this to you?	**0.72**	0.22	0.31	-0.09
01c[e]. Open-ended question: Can you tell us more about this?				
02a. Do you think that other people look at your child because of their atypical genitalia?	**0.64**	0.38	-0.05	0.19
02b. How stressful is this to you?	**0.73**	0.13	0.17	0.21
05a. Do other people speak negatively about *your child* because of their atypical genital or physical appearance?	**0.65**	-0.05	0.27	0.24
05b. How stressful is this to you?	**0.67**	-0.10	0.34	0.14
06a. Do people speak negatively about *you* because of your child?	**0.76**	-0.07	-0.18	0.13
06b. How stressful is this to you?	**0.73**	-0.11	-0.17	0.15
11a. Is your child called names or teased by other children because of their atypical genital or physical appearance?	**0.40**	0.03	0.24	0.55
11b. How stressful is this to you?	**0.41**	0.02	0.44	0.49
Stigma elicited by physical atypicality or cross gender role behaviour [b]				
03a. Can other people see that your child has an atypical physical appearance?	-0.09	**0.76**	0.07	0.35
03b. How stressful is this to you?	-0.14	**0.52**	0.18	0.48
04a. Do you think that other people look at your child because of their atypical physical appearance?	0.39	**0.67**	-0.21	0.16
04b. How stressful is this to you?	0.17	**0.57**	0.13	-0.01
07a. Does your child show more cross-gender role behaviour compared to other children? For parents of daughters: Does your daughter prefer more masculine activities than other girls? For parents of sons: Does your son prefer more feminine activities compared to other boys?	-0.06	**0.87**	0.10	0.20
07b. How stressful is this to you?	-0.01	**0.91**	0.04	0.02

08a. Do other people speak or behave negatively about your child because of their cross-gender role behaviour? (Daughters: masculine behaviour and interests? Sons: feminine behaviour and interests?)	0.11	**0.44**	-0.08	-0.05
Experiences with social exclusion [c]				
09a. Do other people isolate *your child* because of atypical of their genital/physical appearance?	-0.03	0.34	**0.76**	0.19
09b. How stressful is this to you?	0.04	0.24	**0.85**	0.03
10a. Do other people isolate *you* because of your child?	0.17	-0.10	**0.86**	-0.13
10b. How stressful is this to you?	0.21	-0.12	**0.82**	-0.14
12a. Is your child isolated by other children because of their atypical genital or physical appearance?	-0.09	0.00	**0.75**	0.45
12b. How stressful is this to you?	0.02	-0.08	**0.88**	0.22
Emotional reactions [d]				
13a. Does your child suffer from emotional problems because of their atypical genital or physical appearance?	0.26	0.00	-0.07	**0.75**
13b. How stressful is this to you?	0.13	0.02	-0.03	**0.82**
13c. How frequent was your child sad?	0.09	0.06	-0.05	**0.55**
13d. How frequent was your child depressed?	0.01	-0.01	0.07	**0.82**
13g. How frequent was your child shy?	-0.14	0.14	0.13	**0.71**
13h. How frequent was your child socially withdrawn?	-0.11	0.34	0.13	**0.61**
13e. How frequent was your child angry?	0.01	0.19	0.20	0.37
13f. How frequent was your child aggressive?	0.12	0.05	0.28	0.24
14. Are you worried about your child's future?	0.20	-0.01	0.05	0.29
15. Is it difficult for you to accept your child?	0.25	0.07	0.02	-0.12

[a] Stigmatization due to genital ambiguity and stress evoked by such experiences (α = 0.86).
[b] Stigmatization due to atypical physical appearance or displayed cross-gender role behaviour and stress evoked by such experiences (α = 0.84).
[c] Social exclusion or isolation due to DSD and stress evoked by being rejected or isolated (α = 0.88).
[d] Reported emotional problems seen in the child and parental stress evoked these emotional problems (α = 0.85).
[e] Each question 1-12 was followed by an open-ended question: Can you tell us more about this? Example?

SSS-DSD Adult. The PCA extracted three components with Cronbach's alphas ranging between 0.85-0.94. Reliability (internal consistency) of the Adult version was considered as good to very good. The extracted three components explaining 62.9% of the total variance were as follows: a) verbal stigmatization (items 1-2, 4-5, 7; α = 0.92); b) behavioural stigmatization (items 3, 6a, 9-10; α = 0.85); and c) emotional problems due to DSD (items 13-15; α = 0.94). Table 3b shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity of the SSS-DSD Adult was also considered satisfactory.

Table 3*b.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Adult report (*n*=34)

Questions	Components		
	1[a]	2[b]	3[c]
Verbal stigmatisation [a]			
1a. Can other people see that you have genitalia that are (slightly) different from other men/women?	**0.63**	0.03	0.10
1b. How stressful is this to you?	**0.62**	0.36	0.42
1c[d]. Open-ended question: Can you tell us more about this?			
2a. Do you think that other people look at you because of your atypical genitalia?	**0.79**	0.33	0.22
2b. How stressful is this to you?	**0.86**	0.19	0.23
4a. Do you think that other people look at you because of your atypical physical appearance?	**0.71**	-0.08	0.37
4b. How stressful is this to you?	**0.82**	0.21	0.25
5a. Do other people speak negatively about you because of your atypical genital or physical appearance?	**0.75**	0.08	-0.13
5b. How stressful is this to you?	**0.86**	0.10	-0.05
7a. Do other people, including family members, speak or behave negatively about you because you show more cross-gender behaviour compared to others? (For woman: Do you prefer more masculine activities compared to other women? For man: do you prefer more feminine activities compared to other men?)	**0.73**	-0.27	0.12
7b. How stressful is this to you?	**0.71**	-0.23	0.12
Behavioural stigmatisation [b]			
3a. Can other people see that you have an atypical appearance?	0.08	**0.65**	0.41
3b. How stressful is this to you?	0.43	**0.55**	0.23
6a. Do you behave (slightly) differently from other men/women?	0.01	**0.64**	0.17
6b. How stressful is this to you?	0.16	0.36	0.10
9a. Do other people tease you or call you by funny names because of your atypical genital or physical appearance?	0.10	**0.84**	0.07
9b. How stressful is this to you?	0.15	**0.87**	0.18
10a. Do other people isolate/reject you because of your atypical genital or physical appearance?	-0.21	**0.68**	0.23
10b. How stressful is this to you?	-0.21	**0.68**	0.23
Emotional problems [b]			
13a. Do you suffer from emotional problems because of your atypical genital/appearance?	0.31	0.40	**0.75**
13b. How stressful is this to you?	0.31	0.37	**0.75**
13c. How frequently were you sad?	-0.06	0.20	**0.94**
13d. How frequently were you depressed?	0.11	0.16	**0.93**
13e. How frequently were you angry?	0.34	0.31	**0.68**
13g. How frequently were you shy?	0.17	0.16	**0.73**
13h. How frequently were you socially withdrawn?	0.11	0.14	**0.71**
14. Are you worried about your future?	0.21	0.10	**0.74**
15. Is it difficult for you to accept your condition?	-0.02	0.20	**0.75**

[a] Verbal reaction received due to DSD conditions and the stress evoked by such experiences (α = 0.92).
[b] Behavioural reaction received due to DSD conditions and the stress evoked by such experiences (α = 0.85).
[c] Reported emotional problem due to having DSD conditions (α = 0.94).
[d] Each question 1-7,9,10 was followed by an open-ended question: Can you tell us more about this? Example?

Correlations between stigmatization and stress

In both measures, items measuring experiences with stigmatisation were positively and significantly correlated with items measuring stress evoked by such stigmatisation, in all components measured.

SSS-DSD Parent. Stigmatisation due to genital ambiguity positively correlated with stress (r_s (79) = 0.794, $p < 0.001$); stigmatisation elicited by an ambiguous appearance or behaviour positively correlated with stress (r_s (79) = 0.80, $p < 0.001$); social rejection positively correlated with stress (r_s (79) = 0.81, $p < 0.001$); and emotional problems also positively correlated with stress (r_s (79) = 0.64, $p < 0.001$).

SSS-DSD Adult. Verbal stigmatisation positively correlated with stress (r_s (32) = 0.755, $p < 0.001$); behavioural stigmatisation positively correlated with stress (r_s (32) = 0.753, $p < 0.001$); and emotional and acceptance problems due to DSD also positively correlated with stress (r_s (32) = 0.882, $p < 0.001$). The more frequently patients experienced social stigmatisation, the higher their reported stress.

Subgroup analysis

Tables 4a and 4b summarize the comparisons across sex of rearing, treatment status, gender change history (24), and visibility of DSD conditions. In both boys and girls, children and adolescents experienced some degree of stigmatization. Girls reported more stigmatization due to atypical physical appearance or cross-gender role behaviour and had more emotional problems than boys (see Table 4a; gender comparison). Women experienced more stigmatization and had more emotional problems than men. Both men and women experienced some degree of verbal and behavioural reactions due to their DSD conditions (see Table 4b; gender comparison).

Regardless of having received prior hormonal/surgical treatment for DSD, children and adolescents experienced stigmatization and had emotional problems (see Table 4a; treatment status comparison). However, untreated adults experienced more stigmatization than treated adults (see Table 4b; treatment status comparison).

Six youngsters and 15 adults were assigned female at birth but changed gender later in life (24). These patients experienced more stigmatization than patients who kept their initial gender. Young people and adults experienced more stigmatization due to an ambiguous appearance or cross-gender behaviour and had more emotional problems than youngsters who retained the initial sex of rearing (see Table 4a / 4b; social gender role change comparison). Adults who changed gender experienced more behavioural stigmatization than adults who retained the gender assigned at birth (see Table 4b; gender change history comparison).

Children and adolescents with visible ambiguity of the body experienced stigmatization more frequently than patients who could conceal ambiguous characteristics (see Table 4a; visibility of DSD comparison). Regardless of the visibility of DSD, children and adolescents reported emotional problems due to DSD. Adults with visible ambiguity of the body experienced more stigmatization than adults who could conceal ambiguity; this was particularly seen in verbal and behavioural stigmatization (see Table 4b; visibility of DSD comparison).

Table *4* *a.* Median domain and overall sum scores of the SSS-DSD Parent-report across sex of rearing, treatment status, gender change history and visibility of DSD

	Sex of rearing			Treatment status			Gender change history			Visibility of DSD		
SSS-DSD Parent-report	Boys [a] (n=57)	Girls (n=24)	*p* [b]	Treated (n=54)	Untreated (n=27)	*p*	Yes [c] (n=6)	No (n=75)	*p*	Visible (n=12)	Concealable [d] (n=69)	*p*
	median (rar	median (ra		median (ran	median (ran		median (ran	median (ran		median (ran	median (range	
Atypical genitalia score range 10-50	12 [e] (10-50)	10 (10-27)	0.48	11 (10-50)	15 (10-37)	0.20	16 (10-23)	12 (10-50)	0.26	18 (10-32)	11 (10-50)	**0.006**
Atypical appearance / behaviour score range 7-35	10 (10-14)	10 (10-30)	**<0.001**	10 (10-23)	10 (10-30)	0.42	11 (10-30)	10 (10-23)	**0.01**	14 (10-30)	10 (10-21)	**<0.001**
Social exclusion score range 6-30	10 (10-37)	10 (10-23)	0.26	10 (10-37)	10 (10)	0.14	10 (10)	10 (10-37)	0.99	10 (10-37)	10 (10-22)	**0.01**
Emotional problems score range 10-50	10 (10-28)	10 (10-32)	**0.002**	10 (10-32)	10 (10-22)	0.44	14 (10-22)	10 (10-32)	**0.02**	10 (10-32)	10 (10-32)	0.11
Total score [f] score range 33-165	43 (40-103)	49 (40-98)	0.23	43 (40-103)	45 (40-74)	0.47	54 (45-74)	42 (40-103)	**0.02**	61 (40-98)	42 (40-103)	**0.002**

b. Median domain and overall sum scores of the SSS-DSD Adult-report across sex of rearing, treatment status, gender change history and visibility of DSD

	Sex of rearing			Treatment status			Gender change history			Visibility of DSD		
SSS-DSD Adult-report	Men [a] (n=20)	Women (n=14)	*p* [b]	Treated (n=15)	Untreated (n=19)	*p*	Yes [c] (n=15)	No (n=19)	*p*	Visible (n=17)	Concealable [d] (n=17)	*p*
	median (rar	median (ra		median (ran	median (ran		median (ran	median (ran		median (ran	median (range	
Verbal stigmatization score range 10-50	12 [e] (10-47)	10 (10-28)	0.78	10 (10-36)	14 (10-47)	0.14	14 (10-47)	10 (10-28)	0.11	15 (10-47)	10 (10-21)	**0.015**
Behavioural stigmatization score range 8-40	10 (10-26)	11 (10-37)	0.30	10 (10-23)	10 (10-37)	0.25	11 (10-31)	10 (10-37)	**0.03**	13 (10-37)	10 (10-11)	**0.001**
Emotional problems score range 9-45	14 (10-40)	29 (10-47)	**0.009**	12 (10-40)	21 (10-47)	0.07	16 (10-47)	10 (10-44)	0.52	16 (10-47)	16 (10-40)	0.60
Total score [f] score range 17-135	37 (30-91)	50 (31-100)	**0.042**	36 (30-83)	48 (31-100)	**0.046**	41 (31-100)	38 (30-97)	0.80	46 (31-100)	38 (30-63)	0.19

[a] The terms men and women are used according to the gender the patient presented himself or herself socially and to us when he or she participated in the study.
[b] The Mann-Whitney U test was applied.
[c] Ediati A. et al. (24).
[d] Visible refers to all those aspects of physical and behavioural atypicality that cannot be hidden in social interaction. Concealable refers to physical atypicality that can be covered by clothes (partly hidden) and typical phenotype (hidden).

[e] For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion or emotional problems.
[f] Unweighted sum score.

Qualitative data

In text analyses, 4 themes were identified that gave insight into characteristics of social stigmatization and related stress: *a.* (correct, incorrect or lack of) knowledge about DSD, *b.* patients' personality and related emotional and behavioural responses, *c.* cultural norms and related social expectancies and *d.* response from the community.

DISCUSSION

Our study revealed that atypical appearance of the genitals and / or body is problematic (14). Stigmatization was most prominent in patients with an atypical physical appearance who could not hide their ambiguity, in untreated adult patients, in patients who changed their social gender, and in females. The more frequently they experienced DSD-related social stigma, the higher their stress. Patients who were able to hide features of body atypicality from others did not report less emotional problems than patients who had visible features of DSD. This suggests that fear and prevention of being stigmatized is as problematic as having experienced stigmatization. From the qualitative data, we observed that a substantial number of patients withdrew themselves from social interactions, such as withdrawal from school and avoiding interaction with neighbours or community members. In Indonesia, those who show variant sex or gender development are often met with a hostile attitude; patients are humiliated and excluded.

Overall, many patients did not give high rates of experienced social stigmatization; however, patients with atypical physical appearance are vulnerable to social stigmatization. They indicated that stigmatization was stressful, elicited negative emotions, hampered social participation and hence affected overall psychosocial wellbeing. Part of their social stigmatization was related to lack of knowledge about DSD among patients themselves and among Indonesian laymen. We propose that stigmatization can be prevented or reduced by education. Similar to many other non-western countries, Indonesia has few well-trained

medical psychologists available for counselling to help patients and parents cope with DSD. Once educated and supported, patients and parents can then educate their social network to improve their position in the community (30). In addition, educated patients and parents will be better able to decide which treatments are optimal for their particular circumstances.

Indonesia is a collective society in which procreation and progeny are highly valued. Some people with DSD cannot meet such expectations (14,24,25). Our findings are in line with previous studies reporting sexual distress, disclosure dilemmas, and tendency to avoid romantic relationships among women with DSD (25). Women with DSD report a more vulnerable position than affected men in this culture. This may explain why we recruited more male patients (59%) than female patients (41%) for this study. This study includes 20 patients who underwent a female-to-male social gender change, 4 patients changed gender in childhood, 16 of them initiated a change in adolescence or adulthood. Three patients had a 46, XX karyotype and CAH, 17 patients had a 46, XY karyotype (24).

Progressive masculinization may have induced gender dysphoria and instigated the wish to change gender, but ostracism may also contribute to this change.

Limited assessment of the construct validity of the SSS-DSD is a study limitation. Our study focussed on the relationship between social stigmatization and atypical appearance resulting from the delay of medical and surgical treatment. As no suitable measure was available, we developed one. In developing a measure, it is preferred to perform cross-validation studies in addition to principal component analysis to assess construct validity more extensively. Unfortunately, quantitative measures to assess different aspects of psychosocial wellbeing are unavailable in Indonesia and we were unable to perform such analyses (14,24,25).

This study is relevant for patients with DSD who face delay in treatment due to poor understanding of their medical condition, inadequate laboratory support and lack of appropriate and affordable medications (21). Although Western culture is individual-centered

and the demands to follow social norms (e.g. giving birth for females) are less stringent, Western patients with DSD have a vulnerable position in society too. Thus, the current results may be informative to patients and families outside of Indonesia. Ultimately, we aim to optimize patients' psychosexual and psychosocial wellbeing and are searching for adaptations in clinical management that are evidence-based, such as the reduction of stigmatization of those affected by DSD (13).

CONCLUSION

Patients with DSD, particularly those with an atypical appearance, are prone to stigmatization. Such stigmatization is stressful and leads to negative emotional reactions and social isolation. These findings support the assumption that an atypical physical appearance can be harmful for psychosocial wellbeing. This may be particularly true when the medical condition is not understood by the patient, the parents and members of the community, as well as when the patient cannot make their own decisions regarding clinical management. We assume that culturally sensitive education about DSD that is accessible to patients and laymen may remove barriers for social acceptance.

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Authors' Contributions: SMHF and SD initiated the study. SMHF, SD and AW had been involved in written revisions of the manuscript, AE, AD and EB designed the study, analysed the data, produced the figures and performed literature searches and written revisions. AE, JO and AD developed the questionnaires, AE collected the data.

Availability of data and materials

We do not wish to share data originating from our database in order to protect the anonymity of subjects included in this survey. Permission has not been obtained to share data widely with other investigators and would require individual content/assent.

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443 Box

What this known about the subject

- DSD is a somatically and socially challenging condition; many patients and parents suffer from emotional problems, experience or anticipate social stigmatization
- Opposing opinions rule the debate on how to strengthen patients' emotional wellbeing and improve their psychosocial opportunities
- At present DSD-associated social stigma has not been investigated systematically. Such studies are necessary in order to make proper adjustments in clinical management

What this paper adds

- We developed the Social Stigmatization Scale for DSD and investigated patients' and parents' experienced stigma
- Experienced and anticipated DSD related stigmatization was highest among patients with body atypicality and patients who changed gender
- Social stigmatization was evaluated as stressful, related to (self)isolation and highly correlated with depression.

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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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Social stigmatization in late identified patients with disorders of sex development in Indonesia

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ABSTRACT

Objectives: To assess social stigmatization related to atypical appearance of the body, including, but not limited to the external genitalia, among Indonesian patients with a disorder of sex development (DSD). Until recently, diagnostic evaluation, information about the underlying causes of DSD and treatment options were sparsely available for these patients.

Methods: Eighty-one parents of children and adolescents with DSD (aged 6-17 years), and 34 adult patients with DSD (aged 18-41 years) completed the Social Stigmatization Scale towards DSD (SSS-DSD), an instrument developed to assesses the frequency of stigmatization and the level of stress associated with these experiences. Open-ended questions investigated detailed information on stigmatization as well as parents' and patients' emotional and behavioural reactions to these experiences. Differences in stigmatization were explored across sex of rearing, gender change history, treatment status, and DSD characteristics that could be easily identified by others (e.g. masculinisation of the body in females).

Results: Social stigmatization was reported by patients with atypical appearance of their genitalia, atypical appearance of their body aside from their genitals, among those who displayed cross-gender behaviour and those who changed gender. Among participants reared as female, and among children and adolescents who changed gender, social stigmatization was associated with ostracism, depressive symptoms and social isolation.

Conclusions: Patients unable to conceal their condition (those with visible physical atypicality and those who changed gender) experienced social stigmatization. Stigmatization was stressful and related to isolation and withdrawal from social interaction. Education about DSD, self-empowerment and medical interventions to prevent atypical physical development may remove barriers to acceptance by others for affected individuals.

KEYWORDS: social stigmatization, DSD, intersexuality, gender atypicality, body atypicality, Indonesia

INTRODUCTION

Disorders of sex development (DSD) refer to a group of congenital conditions in which development of chromosomal, gonadal, or anatomical sex is atypical, often leading to an atypical appearance of the genitals and other parts of the body that differ in appearance between males and females(1). Clinicians specialised in DSD treatment are confronted with parents' and patients' difficulties in coping with the atypical physical development and the derogatory reactions their atypicality may elicit. In addition to treatments necessary for survival, clinical management aims to reduce or prevent physical atypicality and to enable sexual functioning in order to increase the patient's opportunities for social participation. These interventions have been criticized, as they impact the child's life and are often performed without the child's assent or consent. It has been argued that such interventions do not allow for diversity in sex and gender development and are principally conducted to comfort parents or support the gender ideology of society (2-6). As such, there have been calls to stop this practice of medical and surgical intervention (7-9). However, there is a lack of systematic data on DSD-associated stigma among affected individuals who did not receive such interventions (2). Randomized, controlled studies of early gender assignment, genital surgery and hormonal interventions compared to delayed interventions are highly valued (10) but difficult to conduct. Despite criticisms noted above, most parents living in Western countries choose early gender assignment and surgical correction of the atypical genitalia for their children with DSD (11,12). Follow-up studies on quality of life are scarce and findings are inconsistent regarding the risks and benefits of medical intervention (13-15). Finally, the medical literature contains few reports on DSD and social stigmatisation (16-23).

In Indonesia, DSD is not widely known among health practitioners and laymen. Clinical management is challenged by limited diagnostic and treatment facilities. As a result, many patients live with atypical bodies and experience doubts about their gender (24,25).

During outpatient clinic visits, experiences with social stigmatisation were often reported spontaneously by these patients and stimulated many patients and parents to seek medical help. This enabled us to investigate these patients' experiences of living with physical ambiguity and doubts about their gender (24,25), as well as their experience of social stigmatisation.

METHODS

Study design and setting

Experiences with social stigmatization due to DSD were evaluated from adult patients and parents of affected children and adolescents. Data collection was carried out between March 2007 and May 2011. All patients consulted the DSD Team of the Dr. Kariadi Hospital. The study protocol was approved by the board of the ethical committee at the Faculty of Medicine, Diponegoro University, Semarang, Indonesia.

Patients

All patients with a confirmed diagnosis of DSD consulting the DSD Team of the Dr. Kariadi Hospital (26) were invited to participate in the study. Patients and parents received oral and written study information (provided by AZJ) and provided informed consent. Patients with a genital anomaly and additional features suggestive of a dysmorphic syndrome (27), patients with sex chromosome DSD without mosaicism, and patients with DSD and intellectual disabilities (indicated from the child's academic achievements and/or observed by the medical doctor in interaction with the patient) were excluded. Thirty-four adults (20 men; 14 women; aged 18-41 years) and 81 parents of 60 children (42 boys, 18 girls; aged 6-11) and

21 adolescents (15 boys; 6 girls; aged 12-17 years) participated, with a participation rate of 78%. Table 1 summarizes patient characteristics and diagnoses.

Table 1. DSD diagnoses of participants in the study (N=115)

DSD diagnosis		Age			
		6-11	12-17	18+	Total
Sex chromosome DSD	Patients with 45X/46XY; 46XidicY; 46,XX/46,XY; 46,XX/47,XXY	6	3	5	14
46 XY DSD	AIS*	5	5	6	16
	Gonadal dysgenesis †	6	2	10	18
	Hypomasculinization¥	25	9	7	41
46 XX DSD	CAH – SV‡	18	2	4	24
	Gonadal dysgenesis †	-	-	1	1
	Cloacal malformation	-	-	1	1
Total		60	21	34	115

* Androgen Insensitivity syndrome. AR gene mutation was confirmed (26).
† Abnormal hormonal testicular function with uni/bilaterally undescended testes. The clinical and biochemical presentation suggest gonadal dysfunction. Serum levels of luteinizing hormone and follicle stimulating hormone were elevated but testosterone, anti-müllerian hormone and Inhibin are low for age, and no or diminished serum testosterone response to HCG.
¥ 46 XY karyotype with hypomasculinization of unknown cause, despite extensive analysis (26)
‡ Simple virilising type of congenital adrenal hyperplasia. CYP 21 mutation was confirmed (26).
Details on diagnosis and degree of masculinization at admission per patient can be found in Ediati. et al. (14,24)

Procedure

After obtaining written, informed consent, psychological assessment including data on patients' socio-economic and ethnic-cultural background (14,24,25) was collected in the hospital or at the patient's home, by a trained psychologist (AE).

Materials

Prior to this study, no measure was available to assess social stigmatization in patients with DSD. Therefore, we developed the Social Stigmatization Scale for DSD (SSS-DSD). The SSS-DSD assesses the frequency of experienced stigmatization (1-13a. questions) and

the level of stress evoked by the stigmatizing experiences (1-13b. questions) using a Likert scale with responses ranging from '*not at all*' (1) to '*very much*' (5). In addition, we asked patients to give details about their experiences with DSD, their beliefs on the cause of their DSD, their concerns and ability to cope with DSD (1-12c. questions). We developed parent and adult versions of the SSS-DSD.

The applicability of the SSS-DSD was tested (by AE) prior to implementation and revealed that applying the measure as a paper-pencil test was feasible for well-educated subjects. The rating scale was piloted in a small group of 20 patients and parents with DSD. After a few adaptations, the SSS-DSD seemed suitable for application in this study. Formal large scale psychometric pretesting among sizable numbers of patients or their parents was considered unfeasible in view of the limited numbers of patients with rare genetic conditions. For parents and patients who could not read well, the measure was applied verbally.

Data analysis

Construct validity of both the adult and parental versions of the SSS-DSD scale was explored using principal component analysis (PCA) with varimax rotation and Kaiser Normalization method. Factors with eigenvalues greater than 1 and items with factor loadings (after rotation) greater than 0.40 were considered acceptable. The reliability of the instrument was evaluated using Cronbach's Alpha as a measure of internal consistency.

The overall and domain sum scores of the SSS-DSD were calculated as the unweighted sum scores of the individual domains and items, respectively. For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion and emotional problems. With Spearman's correlation coefficient (*rho*) the correlations between different types of stigma and evoked stress were evaluated. The Kruskal-Wallis test was applied to test for differences in continuous data of more than two groups, the Mann-Whitney

U test for differences between two independent groups. Differences in categorical data were compared using Fisher's Exact test. Differences were considered significant at $p < .05$ (two-sided).

Qualitative data collected were analysed by inductive content analyses using NVivo qualitative data analysis software (28,29). AE started an open coding procedure and finally clustered codes into 4 themes. Relationships between themes were investigated using the compound coding application in NVivo (28,29).

RESULTS

The majority of participants were male, lived in rural areas, Javanese and Muslim. Parents' educational backgrounds varied from no formal education to university level, and the majority attended high school and worked in the lower-income sector or were unemployed. Details on socio-economic and ethnic-cultural variables can be found in Table 2.

Table 2. Participant characteristics (N=115)

Characteristics	Children and adolescents (*n*=81)	Adults (*n*=34)
Gender (of patients)		
Male	57 (70)	20 (59)
Female	24 (30)	14 (41)
Treatment		
Received treatment [a]	44 (54)	15 (44)
No treatment	37 (46)	19 (56)
Social gender role change		
Yes	7 (9)	15 (44)
No	74 (91)	19 (56)
Visibility of DSD [b]		
Visible	12 (15)	17 (50)
Partly hidden	57 (70)	17 (50)
Hidden	12 (15)	
Region		
Central Java	70 (86)	29 (85)
Other provinces in Java	8 (10)	2 (6)
Outside Java island	3 (4)	3 (9)
Ethnic		
Javanese	76 (94)	31 (91)
Non Javanese	5 (6)	3 (9)
Religion		
Islam	77 (95)	33 (97)
Non Islam	4 (5)	1 (3)
Residential setting		
Rural	45 (56)	15 (44)
Suburban	24 (30)	11 (32)
Urban	12 (15)	8 (24)
Highest education attained	(Fathers* / Mothers*)	(Adults)
No formal education	9 (11) / 10 (13)	4 (12)
Elementary school	27 (34) / 28 (35)	3 (9)
High school	36 (45) / 36 (45)	23 (68)
University	8 (10) / 6 (8)	4 (12)
Parents' occupation	(Fathers* / Mothers*)	(Adults)
Unemployed	0 / 44 (55)	13 (38)
Labour	47 (59) / 22 (28)	9 (27)
Self employed	16 (20) / 6 (8)	4 (12)
Staff	17 (21) / 8 (10)	8 (24)

Data are presented in *n* (%) * One father/mother missing for being deceased.

[a] Treatment in most patients had been minimal, for instance, patients had taken glucocorticoid therapy for only a limited period or had undergone one surgical procedure for hypospadias correction when two or more procedures were recommended (14, 25-27)

[b] Social gender role change could be physician imposed, parent imposed or patient initiated (25)

[c] Visibility of DSD refer to all those aspects of physical and behavioural atypicality that cannot be hidden during social interaction. Concealable refers to physical atypicality that can be covered by clothes (partly hidden) and non-ambiguous phenotype (hidden).

Reliability and validity of SSS-DSD parent and adult versions

SSS-DSD Parent. The PCA extracted four components with Cronbach's alphas ranging between 0.84-0.88. Reliability (internal consistency) of the Parent version can be considered as good. The four components explaining 56% of the total variance were as follows: a) stigmatization elicited by genital ambiguity (items 1-2, 5-6, 11; α = 0.86); b) stigmatization elicited by atypical physical appearance or cross-gender role behaviour (items 3-4, 7-8a; α = 0.84); c) social exclusion (items 9-10, 12; α = 0.88); and d) emotional problems due to DSD (items 13a-d, 13g-h; α = 0.85). Table 3a shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity of the SS-DSD Parent was considered satisfactory.

Table 3*a.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Parental report (*n*=81)

Questions	Components			
	1[a]	2[b]	3[c]	4[d]
Stigma elicited by genital atypicality [a]				
01a. Can other people see that your child has genitals that are (slightly) different from that of other children?	**0.60**	0.44	0.07	-0.14
01b. How stressful is this to you?	**0.72**	0.22	0.31	-0.09
01c[e]. Open-ended question: Can you tell us more about this?				
02a. Do you think that other people look at your child because of their atypical genitalia?	**0.64**	0.38	-0.05	0.19
02b. How stressful is this to you?	**0.73**	0.13	0.17	0.21
05a. Do other people speak negatively about *your child* because of their atypical genital or physical appearance?	**0.65**	-0.05	0.27	0.24
05b. How stressful is this to you?	**0.67**	-0.10	0.34	0.14
06a. Do people speak negatively about *you* because of your child?	**0.76**	-0.07	-0.18	0.13
06b. How stressful is this to you?	**0.73**	-0.11	-0.17	0.15
11a. Is your child called names or teased by other children because of their atypical genital or physical appearance?	**0.40**	0.03	0.24	0.55
11b. How stressful is this to you?	**0.41**	0.02	0.44	0.49
Stigma elicited by physical atypicality or cross gender role behaviour [b]				
03a. Can other people see that your child has an atypical physical appearance?	-0.09	**0.76**	0.07	0.35
03b. How stressful is this to you?	-0.14	**0.52**	0.18	0.48
04a. Do you think that other people look at your child because of their atypical physical appearance?	0.39	**0.67**	-0.21	0.16
04b. How stressful is this to you?	0.17	**0.57**	0.13	-0.01
07a. Does your child show more cross-gender role behaviour compared to other children? For parents of daughters: Does your daughter prefer more masculine activities than other girls? For parents of sons: Does your son prefer more feminine activities compared to other boys?	-0.06	**0.87**	0.10	0.20
07b. How stressful is this to you?	-0.01	**0.91**	0.04	0.02

08a. Do other people speak or behave negatively about your child because of their cross-gender role behaviour? (Daughters: masculine behaviour and interests? Sons: feminine behaviour and interests?)	0.11	**0.44**	-0.08	-0.05
Experiences with social exclusion [c]				
09a. Do other people isolate *your child* because of atypical of their genital/physical appearance?	-0.03	0.34	**0.76**	0.19
09b. How stressful is this to you?	0.04	0.24	**0.85**	0.03
10a. Do other people isolate *you* because of your child?	0.17	-0.10	**0.86**	-0.13
10b. How stressful is this to you?	0.21	-0.12	**0.82**	-0.14
12a. Is your child isolated by other children because of their atypical genital or physical appearance?	-0.09	0.00	**0.75**	0.45
12b. How stressful is this to you?	0.02	-0.08	**0.88**	0.22
Emotional reactions [d]				
13a. Does your child suffer from emotional problems because of their atypical genital or physical appearance?	0.26	0.00	-0.07	**0.75**
13b. How stressful is this to you?	0.13	0.02	-0.03	**0.82**
13c. How frequent was your child sad?	0.09	0.06	-0.05	**0.55**
13d. How frequent was your child depressed?	0.01	-0.01	0.07	**0.82**
13g. How frequent was your child shy?	-0.14	0.14	0.13	**0.71**
13h. How frequent was your child socially withdrawn?	-0.11	0.34	0.13	**0.61**
13e. How frequent was your child angry?	0.01	0.19	0.20	0.37
13f. How frequent was your child aggressive?	0.12	0.05	0.28	0.24
14. Are you worried about your child's future?	0.20	-0.01	0.05	0.29
15. Is it difficult for you to accept your child?	0.25	0.07	0.02	-0.12

[a] Stigmatization due to genital ambiguity and stress evoked by such experiences (α = 0.86).
[b] Stigmatization due to atypical physical appearance or displayed cross-gender role behaviour and stress evoked by such experiences (α = 0.84).
[c] Social exclusion or isolation due to DSD and stress evoked by being rejected or isolated (α = 0.88).
[d] Reported emotional problems seen in the child and parental stress evoked these emotional problems (α = 0.85).
[e] Each question 1-12 was followed by an open-ended question: Can you tell us more about this? Example?

SSS-DSD Adult. The PCA extracted three components with Cronbach's alphas ranging between 0.85-0.94. Reliability (internal consistency) of the Adult version was considered as good to very good. The extracted three components explaining 62.9% of the total variance were as follows: a) verbal stigmatization (items 1-2, 4-5, 7; α = 0.92); b) behavioural stigmatization (items 3, 6a, 9-10; α = 0.85); and c) emotional problems due to DSD (items 13-15; α = 0.94). Table 3b shows the factor loadings after varimax rotation and the Cronbach's alpha of each component. The construct validity of the SSS-DSD Adult was also considered satisfactory.

Table 3*b.* Factor loadings after varimax rotation and Cronbach's alphas of the SSS-DSD Adult report (*n*=34)

Questions	Components		
	1[a]	2[b]	3[c]
Verbal stigmatisation [a]			
1a. Can other people see that you have genitalia that are (slightly) different from other men/women?	**0.63**	0.03	0.10
1b. How stressful is this to you?	**0.62**	0.36	0.42
1c[d]. Open-ended question: Can you tell us more about this?			
2a. Do you think that other people look at you because of your atypical genitalia?	**0.79**	0.33	0.22
2b. How stressful is this to you?	**0.86**	0.19	0.23
4a. Do you think that other people look at you because of your atypical physical appearance?	**0.71**	-0.08	0.37
4b. How stressful is this to you?	**0.82**	0.21	0.25
5a. Do other people speak negatively about you because of your atypical genital or physical appearance?	**0.75**	0.08	-0.13
5b. How stressful is this to you?	**0.86**	0.10	-0.05
7a. Do other people, including family members, speak or behave negatively about you because you show more cross-gender behaviour compared to others? (For woman: Do you prefer more masculine activities compared to other women? For man: do you prefer more feminine activities compared to other men?)	**0.73**	-0.27	0.12
7b. How stressful is this to you?	**0.71**	-0.23	0.12
Behavioural stigmatisation [b]			
3a. Can other people see that you have an atypical appearance?	0.08	**0.65**	0.41
3b. How stressful is this to you?	0.43	**0.55**	0.23
6a. Do you behave (slightly) differently from other men/women?	0.01	**0.64**	0.17
6b. How stressful is this to you?	0.16	0.36	0.10
9a. Do other people tease you or call you by funny names because of your atypical genital or physical appearance?	0.10	**0.84**	0.07
9b. How stressful is this to you?	0.15	**0.87**	0.18
10a. Do other people isolate/reject you because of your atypical genital or physical appearance?	-0.21	**0.68**	0.23
10b. How stressful is this to you?	-0.21	**0.68**	0.23
Emotional problems [b]			
13a. Do you suffer from emotional problems because of your atypical genital/appearance?	0.31	0.40	**0.75**
13b. How stressful is this to you?	0.31	0.37	**0.75**
13c. How frequently were you sad?	-0.06	0.20	**0.94**
13d. How frequently were you depressed?	0.11	0.16	**0.93**
13e. How frequently were you angry?	0.34	0.31	**0.68**
13g. How frequently were you shy?	0.17	0.16	**0.73**
13h. How frequently were you socially withdrawn?	0.11	0.14	**0.71**
14. Are you worried about your future?	0.21	0.10	**0.74**
15. Is it difficult for you to accept your condition?	-0.02	0.20	**0.75**

[a] Verbal reaction received due to DSD conditions and the stress evoked by such experiences (α = 0.92).
[b] Behavioural reaction received due to DSD conditions and the stress evoked by such experiences (α = 0.85).
[c] Reported emotional problem due to having DSD conditions (α = 0.94).
[d] Each question 1-7,9,10 was followed by an open-ended question: Can you tell us more about this? Example?

Correlations between stigmatization and stress

In both measures, items measuring experiences with stigmatisation were positively and significantly correlated with items measuring stress evoked by such stigmatisation, in all components measured.

SSS-DSD Parent. Stigmatisation due to genital ambiguity positively correlated with stress (r_s (79) = 0.794, $p < 0.001$); stigmatisation elicited by an ambiguous appearance or behaviour positively correlated with stress (r_s (79) = 0.80, $p < 0.001$); social rejection positively correlated with stress (r_s (79) = 0.81, $p < 0.001$); and emotional problems also positively correlated with stress (r_s (79) = 0.64, $p < 0.001$).

SSS-DSD Adult. Verbal stigmatisation positively correlated with stress (r_s (32) = 0.755, $p < 0.001$); behavioural stigmatisation positively correlated with stress (r_s (32) = 0.753, $p < 0.001$); and emotional and acceptance problems due to DSD also positively correlated with stress (r_s (32) = 0.882, $p < 0.001$). The more frequently patients experienced social stigmatisation, the higher their reported stress.

Subgroup analysis

Tables 4a and 4b summarize the comparisons across sex of rearing, treatment status, gender change history (24), and visibility of DSD conditions. In both boys and girls, children and adolescents experienced some degree of stigmatization. Girls reported more stigmatization due to atypical physical appearance or cross-gender role behaviour and had more emotional problems than boys (see Table 4a; gender comparison). Women experienced more stigmatization and had more emotional problems than men. Both men and women experienced some degree of verbal and behavioural reactions due to their DSD conditions (see Table 4b; gender comparison).

Regardless of having received prior hormonal/surgical treatment for DSD, children and adolescents experienced stigmatization and had emotional problems (see Table 4a; treatment status comparison). However, untreated adults experienced more stigmatization than treated adults (see Table 4b; treatment status comparison).

Six youngsters and 15 adults were assigned female at birth but changed gender later in life (24). These patients experienced more stigmatization than patients who kept their initial gender. Young people and adults experienced more stigmatization due to an ambiguous appearance or cross-gender behaviour and had more emotional problems than youngsters who retained the initial sex of rearing (see Table 4a / 4b; social gender role change comparison). Adults who changed gender experienced more behavioural stigmatization than adults who retained the gender assigned at birth (see Table 4b; gender change history comparison).

Children and adolescents with visible ambiguity of the body experienced stigmatization more frequently than patients who could conceal ambiguous characteristics (see Table 4a; visibility of DSD comparison). Regardless of the visibility of DSD, children and adolescents reported emotional problems due to DSD. Adults with visible ambiguity of the body experienced more stigmatization than adults who could conceal ambiguity; this was particularly seen in verbal and behavioural stigmatization (see Table 4b; visibility of DSD comparison).

Table *4* *a.* Median domain and overall sum scores of the SSS-DSD Parent-report across sex of rearing, treatment status, gender change history and visibility of DSD

	Sex of rearing			Treatment status			Gender change history			Visibility of DSD		
SSS-DSD Parent-report	Boys [a] (n=57)	Girls (n=24)	*p* [b]	Treated (n=54)	Untreated (n=27)	*p*	Yes [c] (n=6)	No (n=75)	*p*	Visible (n=12)	Concealable [d] (n=69)	*p*
	median (rar	median (ra		median (ran	median (ran		median (ran	median (ran		median (ran	median (range	
Atypical genitalia score range 10-50	12 [e] (10-50)	10 (10-27)	0.48	11 (10-50)	15 (10-37)	0.20	16 (10-23)	12 (10-50)	0.26	18 (10-32)	11 (10-50)	**0.006**
Atypical appearance / behaviour score range 7-35	10 (10-14)	10 (10-30)	**<0.001**	10 (10-23)	10 (10-30)	0.42	11 (10-30)	10 (10-23)	**0.01**	14 (10-30)	10 (10-21)	**<0.001**
Social exclusion score range 6-30	10 (10-37)	10 (10-23)	0.26	10 (10-37)	10 (10)	0.14	10 (10)	10 (10-37)	0.99	10 (10-37)	10 (10-22)	**0.01**
Emotional problems score range 10-50	10 (10-28)	10 (10-32)	**0.002**	10 (10-32)	10 (10-22)	0.44	14 (10-22)	10 (10-32)	**0.02**	10 (10-32)	10 (10-32)	0.11
Total score [f] score range 33-165	43 (40-103)	49 (40-98)	0.23	43 (40-103)	45 (40-74)	0.47	54 (45-74)	42 (40-103)	**0.02**	61 (40-98)	42 (40-103)	**0.002**

b. Median domain and overall sum scores of the SSS-DSD Adult-report across sex of rearing, treatment status, gender change history and visibility of DSD

	Sex of rearing			Treatment status			Gender change history			Visibility of DSD		
SSS-DSD Adult-report	Men [a] (n=20)	Women (n=14)	*p* [b]	Treated (n=15)	Untreated (n=19)	*p*	Yes [c] (n=15)	No (n=19)	*p*	Visible (n=17)	Concealable [d] (n=17)	*p*
	median (rar	median (ra		median (ran	median (ran		median (ran	median (ran		median (ran	median (range	
Verbal stigmatization score range 10-50	12 [e] (10-47)	10 (10-28)	0.78	10 (10-36)	14 (10-47)	0.14	14 (10-47)	10 (10-28)	0.11	15 (10-47)	10 (10-21)	**0.015**
Behavioural stigmatization score range 8-40	10 (10-26)	11 (10-37)	0.30	10 (10-23)	10 (10-37)	0.25	11 (10-31)	10 (10-37)	**0.03**	13 (10-37)	10 (10-11)	**0.001**
Emotional problems score range 9-45	14 (10-40)	29 (10-47)	**0.009**	12 (10-40)	21 (10-47)	0.07	16 (10-47)	10 (10-44)	0.52	16 (10-47)	16 (10-40)	0.60
Total score [f] score range 17-135	37 (30-91)	50 (31-100)	**0.042**	36 (30-83)	48 (31-100)	**0.046**	41 (31-100)	38 (30-97)	0.80	46 (31-100)	38 (30-63)	0.19

[a] The terms men and women are used according to the gender the patient presented himself or herself socially and to us when he or she participated in the study.
[b] The Mann-Whitney U test was applied.
[c] Ediati A. et al. (24).
[d] Visible refers to all those aspects of physical and behavioural atypicality that cannot be hidden in social interaction. Concealable refers to physical atypicality that can be covered by clothes (partly hidden) and typical phenotype (hidden).

[e] For all sum scores, a higher score indicates a relatively higher level of stigma, atypicality, social exclusion or emotional problems.
[f] Unweighted sum score.

Qualitative data

In text analyses, 4 themes were identified that gave insight into characteristics of social stigmatization and related stress: *a.* (correct, incorrect or lack of) knowledge about DSD, *b.* patients' personality and related emotional and behavioural responses, *c.* cultural norms and related social expectancies and *d.* response from the community.

DISCUSSION

Our study revealed that atypical appearance of the genitals and / or body is problematic (14). Stigmatization was most prominent in patients with an atypical physical appearance who could not hide their ambiguity, in untreated adult patients, in patients who changed their social gender, and in females. The more frequently they experienced DSD-related social stigma, the higher their stress. Patients who were able to hide features of body atypicality from others did not report less emotional problems than patients who had visible features of DSD. This suggests that fear and prevention of being stigmatized is as problematic as having experienced stigmatization. From the qualitative data, we observed that a substantial number of patients withdrew themselves from social interactions, such as withdrawal from school and avoiding interaction with neighbours or community members. In Indonesia, those who show variant sex or gender development are often met with a hostile attitude; patients are humiliated and excluded.

Overall, many patients did not give high rates of experienced social stigmatization; however, patients with atypical physical appearance are vulnerable to social stigmatization. They indicated that stigmatization was stressful, elicited negative emotions, hampered social participation and hence affected overall psychosocial wellbeing. Part of their social stigmatization was related to lack of knowledge about DSD among patients themselves and among Indonesian laymen. We propose that stigmatization can be prevented or reduced by education. Similar to many other non-western countries, Indonesia has few well-trained

medical psychologists available for counselling to help patients and parents cope with DSD. Once educated and supported, patients and parents can then educate their social network to improve their position in the community (30). In addition, educated patients and parents will be better able to decide which treatments are optimal for their particular circumstances.

Indonesia is a collective society in which procreation and progeny are highly valued. Some people with DSD cannot meet such expectations (14,24,25). Our findings are in line with previous studies reporting sexual distress, disclosure dilemmas, and tendency to avoid romantic relationships among women with DSD (25). Women with DSD report a more vulnerable position than affected men in this culture. This may explain why we recruited more male patients (59%) than female patients (41%) for this study. This study includes 20 patients who underwent a female-to-male social gender change, 4 patients changed gender in childhood, 16 of them initiated a change in adolescence or adulthood. Three patients had a 46, XX karyotype and CAH, 17 patients had a 46, XY karyotype (24).

Progressive masculinization may have induced gender dysphoria and instigated the wish to change gender, but ostracism may also contribute to this change.

Limited assessment of the construct validity of the SSS-DSD is a study limitation. Our study focussed on the relationship between social stigmatization and atypical appearance resulting from the delay of medical and surgical treatment. As no suitable measure was available, we developed one. In developing a measure, it is preferred to perform cross-validation studies in addition to principal component analysis to assess construct validity more extensively. Unfortunately, quantitative measures to assess different aspects of psychosocial wellbeing are unavailable in Indonesia and we were unable to perform such analyses (14,24,25).

This study is relevant for patients with DSD who face delay in treatment due to poor understanding of their medical condition, inadequate laboratory support and lack of appropriate and affordable medications (21). Although Western culture is individual-centered

and the demands to follow social norms (e.g. giving birth for females) are less stringent, Western patients with DSD have a vulnerable position in society too. Thus, the current results may be informative to patients and families outside of Indonesia. Ultimately, we aim to optimize patients' psychosexual and psychosocial wellbeing and are searching for adaptations in clinical management that are evidence-based, such as the reduction of stigmatization of those affected by DSD (13).

CONCLUSION

Patients with DSD, particularly those with an atypical appearance, are prone to stigmatization. Such stigmatization is stressful and leads to negative emotional reactions and social isolation. These findings support the assumption that an atypical physical appearance can be harmful for psychosocial wellbeing. This may be particularly true when the medical condition is not understood by the patient, the parents and members of the community, as well as when the patient cannot make their own decisions regarding clinical management. Culturally sensitive education about DSD that is accessible to patients, families and the community would go a long way towards improving social acceptance and thereby the wellbeing of (young) people with DSD.

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Authors' Contributions: SMHF and SD initiated the study. SMHF, SD and AW had been involved in written revisions of the manuscript, AE, AD and EB designed the study, analysed the data, produced the figures and performed literature searches and written revisions. AE, JO and AD developed the questionnaires, AE collected the data.

Availability of data and materials

We do not wish to share data originating from our database in order to protect the anonymity of subjects included in this survey. Permission has not been obtained to share data widely with other investigators and would require individual content/assent.

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444 Box

What this known about the subject

- DSD is a somatically and socially challenging condition; many patients and parents suffer from emotional problems, experience or anticipate social stigmatization
- Opposing opinions rule the debate on how to strengthen patients' emotional wellbeing and improve their psychosocial opportunities
- At present DSD-associated social stigma has not been investigated systematically. Such studies are necessary in order to make proper adjustments in clinical management

What this paper adds

- We developed the Social Stigmatization Scale for DSD and investigated patients' and parents' experienced stigma
- Experienced and anticipated DSD related stigmatization was highest among patients with body atypicality and patients who changed gender
- Social stigmatization was evaluated as stressful, related to (self)isolation and highly correlated with depression.