

# Subjective need for psychological support (PsySupp) in parents of children and adolescents with disorders of sex development (dsd)

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**Abstract** Disorders/diversity of sex development (dsd) is an umbrella term for congenital conditions often diagnosed within childhood. As most parents are unprepared for this situation, psychological support (PsySupp) is recommended. The aim of this study was to analyse the extent to which parents express a need for PsySupp. Three hundred twenty-nine parents of children with dsd were included; 40.4 % of the parents indicated to have a

need for PsySupp, only 50 % of this group received it adequately. The diagnoses partial gonadal dysgenesis, partial androgen insensitivity syndrome (pAIS) and disorders of androgen synthesis are associated with a high need for PsySupp in parents (54, 65, and 50 %). Sex assignment surgery neither reduced nor increased the need for PsySupp. Taking a picture, radiography, laparoscopy, gonadal biopsy, gonadectomy and hormonal puberty induction are associated with a high need for PsySupp. There was no association between the need for PsySupp and the parents' perception of the appearance of the genitalia.

**Conclusion:** Having a child with dsd is associated with a high need for PsySupp in parents. In particular, parents of children with XY-dsd with androgen effects other than hypospadias expressed a high need of PsySupp. PsySupp for parents should be an obligatory part of interdisciplinary care to reduce fears and concerns.

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## What is known

- In parents, having a child with dsd provokes insecurities and fears. Hence, psychological support is recommended as part of the interdisciplinary care.

## What is new

- This is the first study investigating the subjective need for psychological support in a large sample of parents of children with dsd in Germany. We present data on the subjective need for psychological support of the parents, related diagnoses and factors, which should be considered in psychological counselling.

**Keywords** dsd · Disorders of sex development · Psychological support · Parents · Congenital adrenal hyperplasia

## Abbreviations

CAH Congenital adrenal hyperplasia  
CBAQ Child Behavior and Attitudes Questionnaire

dsd	Disorders/diversity of sex development
dsd-XY-c-f	Diagnostic group with children with female gender of rearing, 46,XY dsd
dsd-XX-p-f	Diagnostic group with children with female gender of rearing with 46,XX dsd and androgen effects
dsd-XY-p-f	Diagnostic group with children with female gender of rearing, 46,XY dsd or mosaicism and partial androgen effects
dsd-XY-p-m	Diagnostic group with children with male gender of rearing, 46,XY dsd or mosaicism and partial androgen effects without androgen effects
pAIS	Partial androgen insensitivity syndrome
cAIS	Complete androgen insensitivity syndrome
PsySupp	Psychological support

## Introduction

“Disorders / diversity of sex development” (dsd) is an umbrella term for rare congenital conditions with incongruence between chromosomal, gonadal and phenotypic sex. Dsd include conditions with diverse genetic aetiology, varying levels of prenatal androgen effects and phenotypes of genitalia and subsequent different medical treatments [11]. The term “dsd” was introduced in 2005 by the Chicago DSD Consensus Group on management of intersex condition to replace nomenclature such as “intersexuality” “hermaphroditism” or “testicular feminization” [8]. According to the Chicago DSD Consensus Group, dsd conditions are classified depending on the karyotype and pathogenesis in three groups: sex chromosome dsd, XY dsd and XX dsd. Sex chromosome dsd include mixed gonadal dysgenesis (46, XY/45, XO), 46XY/46 XX conditions and Turner and Klinefelter syndrome. 46,XY dsd encompass XY gonadal dysgenesis, androgen insensitivity syndrome (AIS), disorders of androgen synthesis and severe hypospadias. 46,XX dsd comprises congenital adrenal hyperplasia (CAH), XX gonadal dysgenesis and uterine and vaginal anomalies. Most parents are unprepared for this complex and challenging situation which can cause confusion, emotional stress, anxiety and perhaps feelings of guilt and shame [14, 16, 25]. Gough et al. highlighted the fundamental shock engendered by the uncertain sex status of children and document parental struggles to negotiate a coherent sex identity for their children [6]. Two recent studies revealed that parents reported overall levels of post-traumatic stress (PTSS) that was comparable to those reported by parents of children diagnosed with cancer [9, 20]. Moreover, another study showed that parents of children with CAH demonstrated symptoms of depression that did not abate with the passage of time [3].

The reaction of parents to the birth of a child with dsd has crucial effects on the atmosphere in which the parent-child

relationship will develop. Parents who also experience caring, accepting and encouraging support from the medical and psychological staff are more likely to be able to develop these attitudes towards their child [23]. An atmosphere of uncertainty, fear and denial hampers this development [22]. Subsequently, dsd of a child can have an impact on the dynamic of the parent-child relationships, the parental and family relationships [9, 24, 26]. The uncertainty, if a decision on the gender of rearing is needed, is especially likely to increase parental stress [10]. Atypical gender role behaviour and gender dysphoria in childhood and adolescence may foster the need for PsySupp in parents.

It has been recommended by the Chicago DSD Consensus Group [8] and the German Ethical Committee [4] that interdisciplinary care of dsd should include a psychologist. On the one hand, the psychologists act as educators and counsellors regarding the psychological aspects of dsd. On the other hand, they have a unique position among all team members facilitating the group process within the team and between the team and family [25]. The appropriate counselling of parents and family starting at diagnosis is needed in order to achieve the best possible development and quality of life of children and adolescents with dsd [7, 18]. This study is a part of the German clinical evaluation study investigating the clinical outcomes and health-care situation of individuals with dsd from 2005–2007 [15]. The aim of this study is to analyse the extent to which parents of children with dsd express a need for PsySupp. To further improve clinical care, factors which are associated with a greater need for PsySupp in parents should be identified.

## Methods

### Study design

The multicenter clinical evaluation study was part of the German network of dsd, funded by the German Ministry of Education and Science (BMBF) [15]. The aim of the study was to describe the clinical outcomes and health-care situation in individuals with dsd. Recruitment took place at four study centres in Germany and at five sites in Austria and Switzerland between January 2005 and December 2007. A psychosocial inquiry of children, adolescents and adults with dsd and their parents was performed. Inclusion criteria of the study followed a generic definition of dsd with laboratory-confirmed diagnosis or clinical diagnosis by a physician. Individuals with Klinefelter or Ullrich-Turner syndrome, severe psychiatric comorbidity and mental disabilities were excluded. Only diagnoses which were formerly known as “intersexuality” and CAH were included. The assessment of the participants and their parents was conducted by trained psychologists during personal encounters. All participants

and their parents gave written informed consent for participation and release of medical data to the interviewer and the principal investigator. A total of 439 children and adolescents, their parents and adults with dsd participated [15].

## Instruments

The study design of the multicenter clinical evaluation study has been described previously [15]. For the substudy, the “dsd-questionnaire” was used: a questionnaire for parents of children with dsd which was developed after consulting experts in the field and reviewing the literature. The dsd questionnaires included sociodemographic variables, questions about medical history, child development, peer relations, and questions concerning dsd (e.g. gender change, knowledge about diagnosis) [11]. Beside the dsd questionnaire, parents were asked to fill out the Child Behavior and Attitudes Questionnaire (CBAQ) [17]. The questionnaire allows for calculating on two scales. The Femininity Scale measuring the extent of typical feminine behaviour (bipolar; high scores=feminine) and the Cross-Gender-Scale measuring the extent of cross-gender behaviour (unipolar; high scores=more cross-gender behaviour) [11].

The need for PsySupp in parents was assessed within the dsd-questionnaire: “Please indicate if you have received psychological counselling/ psychotherapy”. The three categories of response which are related to need for PsySupp in parents were: “We have received psychological counselling/ psychotherapy”, “We have received psychological counselling/ psychotherapy partly”, “We have not received, but we would needed psychological counselling/ psychotherapy”. The answer category “We have not received and we do not need psychological counselling/ psychotherapy” is associated with no need for PsySupp in parents.

Variables that could be related to the need of the parents were selected based on the content from the remaining pool of items, and exploratory data analysis was undertaken.

Diagnoses of children and adolescents were classified into dsd-XX or XY without (c) or with partial (p) androgen effects, and female (f) or male (m) sex of rearing: dsd-XX-p-f, dsd-XY-p-f, dsd-XY-p-m and dsd-XY-c-f. The evaluation was performed on basis of the total cohort and between the diagnostic groups.

## Statistical analysis

For all analyses, SPSS version 22 statistical software was used. The  $\chi^2$  test (2-sided) or, if necessary, the Fisher’s Exact Test and *t* test for independent samples were used to measure associations. *P* values < 0.05 were considered statistically significant.

## Results

### Sample

Caregivers of 329 children and adolescents with dsd (mean age 7.25 years; SD 4.96) participated (Table 1). In 125 cases both parents together, in 185 the mothers, in 14 the fathers and in 4 cases other caregivers answered the questionnaire. In one case, no data as to which caregiver completed the questionnaire was available. Diagnoses of children and adolescents were classified in four diagnostic groups (dsd-XX-p-f, dsd-XY-p-f, dsd-XY-p-m, dsd-XY-c-f) (Table 2) [15].

### Need for PsySupp

One hundred twenty-eight parents (40.4 %) indicated to have a need for PsySupp; 189 parents (59.6 %) reported having no need for PsySupp (Table 3). The need for PsySupp could not be assessed in 12 parents.

### Diagnoses

There was a significant association between the need for PsySupp and the diagnostic groups (*p*=0.010). The need for PsySupp was highest in the group of parents with the child’s diagnosis of dsd-XY-p-f (58.7 %). There was a significant association (*p*=0.025) between the need of PsySupp and the most common diagnoses such as partial/mixed gonadal dysgenesis (54.2 %), pAIS (65 %) and disorders of androgen synthesis (50 %) included in dsd-XY-p-f. The need for

**Table 1** Age, nationality, education level of the parents

	Mothers	Fathers
Total ( <i>n</i> )	315	278
(mean age, SD; range)	(37.3; 6,3; 22–57)	(40.0; 6,9; 20–62)
Nationality		
German	253	249
Austrian	17	18
Swiss	19	19
Two nationalities	2	2
Other	36	31
Total ( <i>n</i> )	327	319
Education level		
Secondary school qualification (without higher education entrance qualification)	169	150
Secondary school qualification (with higher education entrance qualification)	57	46
University degree	82	94
Total ( <i>n</i> ) <sup>a</sup>	308	290

<sup>a</sup> Total numbers are unequal due to missing answers

**Table 2** Description of the cohort: diagnostic groups, diagnoses and age of the children and adolescents with dsd

Group	dsd-XX-p-f <i>n</i> = 132	dsd-XY-p-f <i>n</i> = 66	dsd-XY-p-m <i>n</i> = 108	dsd-XY-c-f <i>n</i> = 22
Definition	Female gender of rearing with 46,XX dsd and androgen effects	Female gender of rearing with 46,XY dsd or mosaicism and partial androgen effects	Male gender of rearing with 46,XY dsd or mosaicism and partial androgen effects	Female gender of rearing with 46,XY dsd without androgen effects
Karyotype	XX	XY+	XY+	XY
Gender of rearing/recent gender	Female	Female	Male	Female
Androgen effects	Present	Present	Present	None
AIS	–	12 pAIS	9 pAIS	12 cAIS
Gonadal dysgenesis	1 partial/mixed gonadal dysgenesis	29 partial/mixed gonadal dysgenesis	21 partial/mixed gonadal dysgenesis	9 complete gonadal dysgenesis
Disorders of androgen synthesis	–	17beta-hydroxysteroid- dehydrogenase III deficiency (7), 5 alpha-reductase II deficiency (4), LH-receptor defect (1), 17/20-lyase deficiency (1)	5alpha-reductase II deficiency (3), LH-receptor defect (3), 17beta hydroxysteroid-dehydrogenase III deficiency (2), not precisely classified (2)	–
CAH	CAH (128)	–	–	–
Severe hypospadias	–	Severe hypospadias (1)	Severe hypospadias (50)	–
Other conditions	Complex malformation (1), aromatase deficiency (2),	Complex malformation (5), penile agenesis (1), cloacal exstrophy (1), unclassified clinical diagnoses of dsd (4)	Disorder of anti-mullerian hormone (1), Complex malformation (3), 46, XX-male(3), micropenis (2), unclassified clinical diagnoses of dsd (9) <sup>a</sup>	Clinically most likely SF1 mutation, but defect was not found (1)
Newborns	8	4	10	1
6 months–3 years	29	7	36	1
4–7 years	29	12	34	5
8–12 years	33	24	21	8
13–16 years	33	19	7	7

Excluded from diagnosis group comparisons: one child with 46,XX and without androgen effects (6 months–3 years)

AIS androgen insensitivity syndrome, CAH congenital adrenal hyperplasia, cAIS complete androgen insensitivity syndrome, dsd disorders/diversity of sex development, pAIS partial androgen insensitivity syndrome, + incl. mosaics with parts of “Y chromosome” (in some cases chromosome status has not been investigated, in these cases classification results according to the clinical status)

<sup>a</sup> Included in XY-dsd-p-m despite karyotype 46,XX

**Table 3** Need for PsySupp in parents

Total of parents		317	%
Need for PsySupp		128	40.4
Divided in:	“We have received psychological counselling/ psychotherapy”	29	(22.7)
	“We have received psychological counselling/ psychotherapy partly”	32	(25.0)
	“We have not received but we needed psychological counselling/ psychotherapy”	67	(52.3)
No need for PsySupp		189	59.6
“We have not received and we do not need psychological counselling/ psychotherapy”			

PsySupp was lowest in parents of children with CAH (33.9 %) or severe hypospadias (26.5 %) (Table 4).

### Parents’ perception of the appearance of the genitalia, gender of rearing and gender role behaviour

No significant association could be found between the need for PsySupp of parents and their perception of the genitalia of the child at birth and gender of rearing (Table 5).

A change of the gender of rearing was performed in 32 children of the diagnostic groups dsd-XX-p-f (male to female  $n=15$ ; diagnosis CAH; mean age of change of gender of rearing 0.30 years), dsd-XY-p-f (male to female  $n=9$ ; diagnosis: partial gonadal dysgenesis  $n=6$ ; PAIS  $n=2$ ; other  $n=1$ ; mean age of change of gender of rearing 0.72 years) and dsd-XY-p-m (female to male  $n=8$ ; diagnosis: partial gonadal dysgenesis  $n=2$ ; severe hypospadias  $n=2$ ; other  $n=4$ ; mean age of change of gender of rearing 0.84 years). The gender change was initiated by doctors in 19 cases (mean age 0.37 years) and by parents in 9 cases (mean age 1.01 years). There was no second change of the gender of rearing in the sample. Sixteen parents (53.3 %) reported a need for PsySupp if there was a change of the gender of rearing ( $p=0.171$ ).

There was an association of need for PsySupp in parents and preferred gender of playmates of the child in the diagnostic group dsd-XX-p-f ( $p=0.038$ ). There was a high need for PsySupp if the children preferred to play with boys (81.8 %) or girls (64.0 %). There was a low need for PsySupp in parents if the children preferred to play with both genders (37.5 %) (Table 6). This was not the case in the other diagnostic groups. Although not significant due to small sample size, a difference may also apply to the diagnostic group dsd-XY-p-m.

Gender role behaviour measured with the CBAQ [11, 12] was also associated with need for PsySupp in parents of children within the diagnostic group dsd-XY-p-f. There was a higher need for PsySupp when the parents indicated that their child showed more cross-gender behaviour (Table 7). This was not case in the other diagnostic groups.

### Surgery, medical investigations and hormonal treatment

No association between the need for PsySupp in parents and genital reconstructive surgery could be found. Genital reconstructive surgeries were: female urethral correction ( $n=27$ , scheduled in  $n=7$ ), male urethral correction ( $n=67$ , scheduled in  $n=18$ ), vaginoplasty ( $n=84$  scheduled in  $n=41$ ), feminising surgery of the clitoris ( $n=102$ , scheduled in  $n=12$ ), vaginal dilation ( $n=11$ , scheduled in  $n=13$ ), testicular relocation ( $n=38$  scheduled in  $n=10$ ) and masculinisation surgery of the penis ( $n=59$  scheduled in  $n=16$ ).

Two hundred seventy (82 %) parents (117 dsd-xx-p-f, 49 dsd-XY-p-f, 98 dsd-XY-p-m, 6 dsd-XY-c-f) indicated that genital surgery of the child was recommended. No association between the need for PsySupp in parents and recommended genital surgery could be found.

In our sample, parents reported different previous medical procedures. An association ( $p=0.027$ ) between gonadectomy ( $n=72$ , scheduled in  $n=9$ ) and need for PsySupp in parents could be found (Table 4).

Parents reported various medical investigations in the context of their child’s diagnosis. There was a significant association between need for PsySupp in parents and medical investigations such as taking a photo ( $p=0.006$ ), radiography ( $p=0.028$ ), laparoscopy ( $p=0.009$ ), and gonadal biopsy ( $p=0.003$ ) (Table 4). No association to need for PsySupp was found for blood sampling ( $n=285$ ), magnetic resonance imaging (MRI;  $n=23$ ), computerised tomography (CT;  $n=18$ ), endoscopy ( $n=43$ ) or ultrasound ( $n=249$ ).

An association between need for PsySupp in parents and previous or planned hormonal induction of puberty could be found ( $p=0.01$ ) (Table 4).

### Understanding of the diagnosis

One hundred nineteen (40.9 %) parents reported that they did not completely understand the information about the final diagnosis; 172 (59.1 %) parents reported that they did understand the information. No association between the need for PsySupp and understanding the diagnosis could be found.

**Table 4** Significant results of the study

	Need for PsySupp		No need for PsySupp		Number
	<i>n</i>	%	<i>n</i>	%	
Total	128	40.4	189	59.6	317
Diagnostic groups**					
dsd-XX-p-f	43	33.6	85	66.4	128
dsd-XY-p-f	37	58.7	26	41.3	63
dsd-XY-p-m	40	38.5	64	61.5	104
dsd-XY-c-f	8	38.1	13	61.9	21
Main diagnoses*					
pAIS	13	65.0	7	35.0	20
cAIS	5	41.7	7	58.3	12
Partial/mixed gonadal dysgenesis	26	54.2	22	45.8	48
complete gonadal dysgenesis	3	37.5	5	62.5	8
Disorders of androgen synthesis	11	50.0	11	50.0	22
CAH	42	33.9	82	66.1	124
Severe hypospadias	13	26.5	36	73.5	49
Other	15	44.1	19	55.9	24
Surgeries					
Gonadectomy*	36	52.2	33	47.8	69
Investigations in context of the diagnosis					
Taking a picture**	70	49.0	73	51.0	143
Radiography*	63	47.7	69	52.3	132
Laparoscopy**	16	66.7	8	33.3	24
Gonadal biopsy**	38	56.7	29	43.3	67
Induction of puberty**					
Hormonal induction of puberty—received	15	50.0	15	50.0	30
Hormonal induction of puberty—planned	28	58.3	20	41.7	48
No hormonal introduction of puberty	63	35.0	117	65.0	180
Nationality of the father**					
German, Austrian, Swiss, two nationalities	105	37.6	174	62.4	279
Other	19	65.5	10	34.5	29
Education level of the mother*					
Secondary school qualification (without higher education entrance qualification)	69	61.7	93	57.4	162
Secondary school qualification (with higher education entrance qualification)	14	25.0	42	75.0	56
University degree	37	47.4	41	52.6	78
Occupation of the father***					
Full-time work	89	35.6	161	64.4	250
Part-time work	8	53.3	7	46.7	15
Unemployed	10	71.4	4	28.6	14
Retired	6	85.7	1	14.3	7

Only significant results are stated: \* $p \leq 0.05$ ; \*\* $p \leq 0.01$ ; \*\*\* $p \leq 0.001$  with respect to corresponding crosstab

### Age at diagnosis, duration of making the diagnosis and sources of information

No significant association with the need for PsySupp of parents could be found for the age at first diagnosis (in 26 cases the diagnosis was confirmed before birth, in 240 cases in the

first 5 days after birth, in 39 cases in the first year and in 24 cases after 1 year); the duration of making the diagnosis (in 74 cases making the diagnosis took 1–7 days, in 80 cases 1–4 weeks, in 90 cases 1–12 months and in 20 cases longer than 1 year); for presence of previous case of familial dsd ( $n=39$ ) and for sources of information. In addition to information

**Table 5** Parents' perception of the genitalia and need for PsySupp (not significant)

"How did you assess your child's genitalia immediately after birth?"	Male		Ambiguous		Female	
	<i>n</i>	Need for PsySupp <i>n</i> (%)	<i>n</i>	Need for PsySupp <i>n</i> (%)	<i>n</i>	Need for PsySupp <i>n</i> (%)
dsd-XX-p-f	30	13 (43.3)	34	9 (26.5)	57	16 (28.0)
dsd-XY-p-f	8	5 (62.5)	14	8 (57.1)	40	24 (60.0)
dsd-XY-p-m	72	24 (33.3)	19	8 (42.1)	6	9 (66.7)
dsd-XY-c-f	0		0		21	8 (38.1)
Total	110	42 (38.2)	67	25 (37.3)	127	54 (42.5)

about the condition from medical staff, parents used the Internet ( $n=177$ ), books ( $n=107$ ) and/or information from self-help groups ( $n=104$ ).

### Sociodemographics

Significant associations between need for PsySupp in parents and nationality of the father ( $p=0.005$ ), level of education of the mothers ( $p=0.024$ ) and unemployment of the father ( $p=0.001$ ) could be found (Table 4).

No significant association could be found between the need of PsySupp and who responded the questionnaire, age of the parents, age of the child, importance of religion, persuasion, size of the home town and number of siblings.

### Free comments of the parents

In addition to the quantitative questions, the parents could express their views and opinions as free comments. Some qualitative impressions of desires and needs of the parents with need for PsySupp were: "I needed somebody giving me confidence and information", "in the first years I strongly desired psychological care", "Open conversation with hospital staff", "a cure", "Psychological guidance for the development of my child", "psychological support at diagnosis immediately after birth", "contact and exchange with other parents", "That I was perceived", "someone who encourages

me and stands by me during the period of uncertainty immediately after birth and in the first weeks", "I would like to have a contact person to get some advice for upcoming problems and fears in the future", "Support within the family", "Counselling about the future, puberty; e.g. medical examinations, advice on possible hormonal treatments, possible problems and how to be prepared for them", "to be able to talk about concerns and fears before surgery", "a better comprehension of the physicians", "during the first surgery I should have had help, but I was much too focused on my child to ask"

### Discussion

The study shows that parents of children with dsd have a high subjective need for PsySupp (40.4 %). However, only about half of the parents with need for PsySupp received it adequately or partly and half needed it but did not receive any PsySupp. We assume that, in the twenty-first century, the services offering professional PsySupp in children's hospitals were not well developed in Germany. Leidolf et al. reported in 2008 that 69 % of 29 paediatric endocrinology fellowship training offered PsySupp and 58 % had a mental health specialist on staff, but only 19 % of patients or families received emotional support at diagnosis and only 15 % of patients or families after diagnosis [13]. Pasterski et al. found that 95 % of centres in

**Table 6** Preferred gender in friendships and need for PsySupp in parents

"Who are currently your child's friends?"	Girls		Boys		Both		Unknown	
	<i>n</i>	Need for PsySupp (%)	<i>n</i>	Need for PsySupp (%)	<i>n</i>	Need for PsySupp (%)	<i>n</i>	Need for PsySupp
dsd-XX-p-f	37	15 (40.5)	25	8 (32.0)	42	13 (31.0)	4	2
dsd-XY-p-f*	25	16 (64.0)	11	9 (81.8)	16	6 (37.5)	2	0
dsd-XY-p-m	7	5 (71.4)	35	13 (37.1)	37	13 (35.1)	3	0
dsd-XY-c-f	13	4 (30.8)	1	0	6	3 (50.0)	0	0
Total	82	40 (48.8)	72	30 (41.7)	102	35 (34.3)	9	3

\*Significant  $p \leq 0.05$

**Table 7** CBAQ and need for PsySupp in parents; diagnostic group dsd-xy-p-f

	Need for PsySupp	No need for PsySupp
<i>n</i> (%)	21 (65.6)	11 (34.4)
Femininity scale* Mean (SD)	63.3 (8.8)	72.6 (7.5)
Cross-gender scale** Mean (SD)	26.6 (7.6)	18.4 (4.1)

Femininity scale measuring the extent of typical feminine behaviour (bipolar; high scores=feminine); Cross-Gender-Scale measuring the extent of cross-gender behaviour (unipolar; high scores=more cross-gender behaviour); only significant results are stated: \* $p \leq 0.01$ ; \*\* $p \leq 0.001$

Europe treating children with dsd in 2010 offered primary psychological support services such as a child psychiatrist or psychologist [21]. Moreover, Pasterski et al. found that the average uptake of PsySupp services by parents of a child with dsd was only 54 %, which is higher than the need of for PsySupp in parents in this study (40 %) [21]. However, no data are available how often PsySupp was offered in this study. Subsequently, potential average uptake cannot be evaluated. Although only 40 % of the parents indicated a subjective need for PsySupp, a higher real need for PsySupp in parents might exist from a psychotherapeutic perspective. We assume that fears of stigmatisation by psychotherapy and psychological counselling or ignorance about the benefit, content or aims of the different forms of PsySupp are still factors for parents not accepting PsySupp or for physicians not offering it. Additionally, parents without offered PsySupp might not be aware of their needs.

These studies show that the recommendations of the Chicago consensus 2005, to involve a psychologist in the care of a child with dsd, have been mostly implemented now in Europe. Thus, parents with need for PsySupp have better access to it than before 2005.

## Diagnoses

The highest need for PsySupp (58.7 %) was reported by parents of children with conditions included in the diagnostic group dsd-XY-p-f. The diagnoses of partial gonadal dysgenesis, partial androgen insensitivity syndrome (pAIS) and disorders of androgen synthesis, which are included in the diagnostic group dsd-XY-p-f and dsd-XY-p-m, are particularly associated with a high need for PsySupp in parents (54.2, 65.0 and 50.0 %). This high need of PsySupp in parents of children with these conditions can be explained by the uncertainties about the sex of the child where a decision about the gender of rearing is needed. In contrast, parents having a child with severe hypospadias or CAH (46,XX) had much less need for PsySupp (33.9 and 26.5 %). This difference can be explained by the more consistent assignment to male or female gender of

rearing. Individuals with CAH (46,XX) are primarily seen as girls and individuals with severe hypospadias (46,XY) as boys. This association is not only related to the appearance of the external genitalia but also to the internal genitalia and possible future fertility. The unambiguity of the gender of rearing reduces uncertainties and confusion and apparently simplifies dealing with dsd for parents and for the medical staff because the question, “Is it a boy or a girl?” can be answered [6].

## Surgery, medical investigations and hormonal treatments

Our data show that sex assignment surgery neither reduces nor increases the need for PsySupp in parents. Schober argues that surgery makes parents and doctors more comfortable, but counselling makes people comfortable too, and it is not irreversible [14, 27]. Crissman et al. point out that immediate surgery reduces early parental concerns regarding genital appearance, but does not eliminate worries about the child’s future gender development or sexual functioning [1]. As surgery does not reduce the need for PsySupp in parents, the fears and concerns of parents should not be the reasons for sex assignment surgery. It is therefore important to keep in mind that parents want to do the “right thing” [19]. Moreover, parental decisions concerning early sex assignment surgery depend highly on the medical advice. But neither physicians nor parents are fully aware of the magnitude of this medically induced influence [28] and may later be burdened with decision regret.

Investigations in context of the diagnosis such as taking a photo, radiography, laparoscopy and gonadal biopsy are associated with a great need for PsySupp in parents (49.0, 47.7, 66.7, and 56.7 %). Consequently, only those interventions specifically indicated for further treatment planning should be performed. To reduce their stress and concerns, parents should always be informed in an adequate and understandable manner about the aims and procedures of the different medical investigations. Taking a picture might be associated with need for PsySupp in parents because of stress to them from pointing out the unusual nature of the child’s genitalia. Taking a picture should therefore not be undertaken unless absolutely necessary for treatment planning. In the medical context, a drawn sketch is often sufficient.

There was a greater need of PsySupp in parents when gonadectomy of the child was performed (52.2 %) and if puberty was induced or was planned to be induced hormonally (50.0 and 58.3 %). Gonadectomy is an irreversible intervention with far-reaching consequences for affected individuals as it results in deficiency of own sex hormone production and necessitates subsequent lifelong hormone substitution. Therefore, it is important that the interdisciplinary team respond to fears and concerns of parents if a gonadectomy is planned. Hormonal induction of puberty should be raised and discussed, if



needed, in the period from childhood to adolescence as, even in early childhood, it can cause insecurity in parents.

### Parents' perception of the appearance of the genitalia, gender of rearing and gender role behaviour

There was no association between parents' perception of the appearance of the child's external genitalia, and need of PsySupp in parents. Duguid et al. and Pasterski et al. also did not find an association between parental post-traumatic stress, self-esteem, psychological stability and genital ambiguity of the child [5, 20]. Therefore, it can be assumed that, in the context of the need for PsySupp in parents, the isolated issue of ambiguous genitalia plays a minor role. It is more likely that the whole complex situation of having a child of uncertain sex and future, without a definite option of "cure", causes distress for parents.

Gender role behaviour and gender specifics in the preferred gender of playmates of the child play a role only in the diagnostic group dsd-xy-p-f. In the other groups, atypical gender behaviour is not associated with need for PsySupp in parents. We assume that this association is connected to the uncertainty of gender in this diagnostic group as already discussed under diagnoses. As the assignment to a male or female gender of rearing of the children is more consistent in the other diagnostic groups, atypical gender behaviour may not confuse parents. If children behave atypically, in cases where there was a difficult decision on gender of rearing, parents could be insecure with respect to this previous decision. This may foster the need for PsySupp in parents.

### Understanding of the diagnoses

Unexpectedly, there was no association between understanding the diagnoses and the need for PsySupp. It can be concluded that, with respect to the need for PsySupp, a precise understanding of the dsd condition is not crucial. However, it appears alarming that 41 % of the parents did not understand the diagnosis completely. This is probably due to the complexity of the individual condition, which even for professionals is often difficult to understand. Nevertheless, parents and patients should be informed adequately and understandably about the dsd condition.

### Sociodemographic factors

We found no association between the age of the children and need for PsySupp in parents. Thus, it can be concluded that PsySupp for parents is relevant from childhood to adulthood. Hullmann et al. confirm that caregivers of children with dsd may have different psychosocial needs depending upon their child's development stage and based upon the disorder-related challenges that are salient at that developmental stage [9].

Despite our sample not including parents directly at the first suspicion of a dsd condition, it is possible that the need for PsySupp in parents is greater at diagnosis than at a later point of time when the parents have adapted to the situation [25]. No distinction between the maternal and paternal need of PsySupp could be performed as parents answered the questionnaire together. Nevertheless, we found associations between maternal and paternal sociodemographic data and the parental need for PsySupp. The need for PsySupp was greater (>65 %) if fathers had non-German citizenship, being part-time employed, unemployed or retired. It is likely that fathers of non-German citizenship might have problems with understanding the dsd condition and treatment. Moreover, different cultural views of dsd and stigmatisation might be factors inducing increased need of PsySupp in these families. In contrast, impaired paternal employment by itself can be a factor for increased need of PsySupp for families. Moreover, differences in the need for PsySupp related to the maternal education level were found. There was a higher need for parental PsySupp when the mother held a university degree. This finding could be explained by a higher uptake of PsySupp due to less stigmatisation of psychological counselling in academics. In contrast, parents with low maternal education level also had a higher need for PsySupp which might be caused by less understanding of dsd and fears of stigmatisation. Studies differentiating between maternal and paternal need of PsySupp in dsd are needed to verify these differences. Some authors particularly highlighted the need for information and education by parents, as dsd is a rare condition and mostly unknown in society [2, 5].

### Limitations of the study

The study was part of a large quantitative cross-sectional outcome study. Consequently, neither quantitative data explaining why such a high proportion of parents did not receive PsySupp nor on the specific needs for PsySupp of dsd families could be collected. However, some qualitative comments of parents were available. Parents expressed the following needs: openness, confidence and information, encouragement, advice how to be prepared for possible problems in the future, contact and exchange with other parents, possibility of talking about fears and concerns before surgery, family support (see results: free comments of the parents).

### Conclusion

Parents of a child with dsd have a high subjective need for PsySupp, but half of them did not receive it as needed. In particular, a high need for PsySupp is indicated for parents of children and adolescents with partial or mixed gonadal dysgenesis, pAIS and disorder of androgen synthesis (children

with XY-dsd with androgen effects). In the last decade, the possibility of receiving PsySupp for families of a child with dsd have improved in Europe but the uptake of PsySupp seems not to be adequate.

PsySupp for parents of children with dsd should be an obligatory part in the interdisciplinary care of children and adolescents to reduce fears of stigmatisation by psychological counselling and to enable parents to provide their children with good parental support. So far, the method, extent and content of PsySupp is based on the individual situation and needs of the parents as yet no special concepts for PsySupp in dsd are available. Interdisciplinary teams with psychologically trained medical staff and medically trained psychologists are needed to reduce fears and concerns regarding gender issues, hormonal treatment and surgery. However, the structure, scope and content of PsySupp for dsd have to be improved and programmes for PsySupp for dsd developed.

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