



Satisfaction with a new patient education program for children, adolescents, and young adults with differences of sex development (DSD) and their parents.

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ABSTRACT

Objective: Evaluation of the participant satisfaction with a newly developed interdisciplinary, modular education program for children, adolescents, and young adults with differences of sex development (DSD) and their parents.

Methods: The two-day program including tailored medical information, peer consultation and psychological support aimed to improve diagnosis-specific knowledge and empowerment. Post-training satisfaction was measured using an adapted ZUF-8 questionnaire, scoring from 5 (worst) to a maximum of 26 (best) for persons aged 6–17 and from 10 to 40 points for adults, including 2 open-ended questions.

Results: The questionnaire, completed by 89 children (6–13 years), 92 adolescents (14–17 years), 47 young adults (18–24 years), and 345 parents, revealed consistent high satisfaction with the program regardless of age or diagnosis (children 24.4 ± 2.1 , adolescents 23.5 ± 2.7 ; young adults 36.0 ± 4.0 , parents 36.6 ± 3.4). Neither sociodemographic factors nor diagnosis burden, shame, or informedness showed relevant associations with satisfaction levels. Participants highlighted exchange and open atmosphere as key satisfaction elements.

Conclusion: Satisfaction with the new education program was high in all examined groups. Implementing it in routine care requires further analysis to determine the program's long-term effects on well-being and knowledge.

Innovation: The first educational program for young people with DSD addressing their specific challenges through inclusive language, an open approach to sex and gender and the inclusion of self-help groups.

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1. Introduction

Differences of sex development (DSD) is a broad “umbrella term” [1,2] covering a large spectrum of rare conditions affecting gonads, adrenal glands, and sex characteristics. According to the DSD classification, diagnoses are classified into 3 groups: 1) chromosomal DSD, e.g. Turner syndrome, Klinefelter syndrome and mosaic forms such as 45,X/46,XY, 2) 46,XX-DSD, e.g. congenital adrenal hyperplasia (CAH) and 3) 46,XY-DSD, e.g. complete androgen insensitivity syndrome [2]. In the following, “differences” is used instead of the common term “disorders” to avoid inadvertently emphasizing disease; rather, it is intended to refer to variances in sex development. Taken together they affect up to 1.7% of children worldwide [3,4] with an incidence of 1:4500–5000 live births, varying depending on the condition [5], impacting puberty, fertility, and sexual health [6]. Some of the DSD entities require lifelong hormone replacement, and conditions like CAH risk life-threatening adrenal crises. In others, self-perceived gender identity may differ from physical appearance. Psychological wellbeing can also be affected, leading to reduced quality of life, increased anxiety and depression, body image issues, and higher likelihood of harassment compared to peers without DSD [7,8]. Healthcare in the past has not always adequately supported positive self-esteem or informed, confident management of the condition [9]. Therefore, since 2006, experts have emphasized the need for psychosocial care alongside medical treatment [2,5,10]. Affected families bring many emotional, social and cultural issues into the processing of the diagnosis, and many lack adequate knowledge about the sexual differentiation process [11].

Patient education programs are standardized, manualized and interactive group programs for patients with chronic diseases taught by qualified health care professionals [12]. They are focused on providing knowledge about the diagnosis and helping patients to cope with the diagnosis in everyday life, at work, or at school. Emphasis is placed on self-management, psychosocial support, and interaction with similarly diagnosed individuals [13]. The overall goal is empowerment of individuals with the condition and their families. In recent years, the importance of patient education programs for chronic diseases has been recognized, and education programs for numerous diagnoses have been developed [14–17]. However, for persons with DSD standardized programs for training and psychosocial support with stringent evaluations are still lacking.

In developing a standardized concept for the education of children, adolescents, young adults with a DSD diagnosis and their parents, one of the challenges is to adapt existing concepts for education on chronic diseases (e.g., ModuS) [18,19]. The aim was to empower participants in dealing with their diagnosis, to provide knowledge on etiology and therapy, while reducing pathologization of the diagnoses covered by the term DSD. Instead, the focus is on the consideration of sex variants as a biological feature.

Within the project Empower-DSD, an educational program for minors and young adults with DSD as well as their parents was developed and evaluated. The aim of the present analysis was to evaluate satisfaction across the different groups of participants (children, adolescents, young adults, parents) as well as for different subgroups (e.g., by diagnosis). Furthermore, factors influencing satisfaction were identified to consider them in future trainings as well as to improve the curricula.

2. Methods

2.1. The DSD education program

Four diagnosis-specific, modular curricula for two-day training sessions each were developed for congenital adrenal hyperplasia (CAH), for Turner syndrome (TS), for Klinefelter syndrome (KS), and for the other DSD diagnoses (remaining chromosomal DSD, 46,XY-DSD, 46,XX-DSD, also including Mayer-Rokitansky-Küster-Hauser (MRKH) syndrome). The development process of the training and the qualification of the

educators has been described by Wiegmann et al. [20]. Separate curricula for each diagnosis and for each group of participants contained defined learning objectives, content, and learning methods for the modules (Fig. 1). The training program consisted of 12–14 lessons of 45 min each, organized in 7 modules on two consecutive days. The courses were held in groups that were kept as homogeneous as possible in terms of age and sex/gender, with the training of parents and children provided in separate, parallel groups. The training courses were developed for a group size of 4–8 participants. Due to the COVID-19 pandemic, smaller groups, the use of masks, social distancing and testing had to be implemented at times in order to be able to continue the training courses in person.

The DSD education program has the following unique characteristics:

- (1) Addressing the specific challenges of people with DSD and connecting general psychosocial phenomena of chronic diseases to their specifics and without pathologizing sex variants.
- (2) Involving self-help groups in a participatory approach during the development, implementation as well as evaluation of the curriculum [21,22].
- (3) Integration of an open attitude towards sex and gender in the curriculum and its realization by the educators with the use of an inclusive language.
- (4) Use of specific materials that emphasize physical diversity (Fig. 2).

2.2. Study design and study population

The evaluation of satisfaction with the educational program was done immediately after it had been completed. Training sessions were conducted at five study centers spread across Germany: Charité - Universitätsmedizin Berlin (1), St. Josefs Hospital Bochum (2), University of Lübeck (3), University Hospital Münster (4), and University Hospital Ulm (5). The investigation is part of the extensive evaluation concept of the Empower-DSD study, conducted between 2019 and 2023, the study protocol of which has been published elsewhere [20]. Children, adolescents, and young adults between 6 and 24 years of age who had one of the following diagnoses confirmed by chromosomal analysis, genetic test result, laboratory test, or clinical examination were included: CAH, KS, TS, and other DSD (remaining chromosomal DSD, 46,XY-DSD, 46,XX-DSD, MRKH syndrome - in the following referred to as “other DSD”). Furthermore, parents, primary caregivers or relatives (hereafter referred to as parents) of children starting from the age of birth with one of the corresponding diagnoses were eligible to participate in the training and evaluation. Parents were categorized into parents of newly diagnosed children who had received their diagnoses within the previous 2 years and parents of children with longstanding diagnoses. Families were excluded if they lacked sufficient German language skills.

A signed informed consent for participation in the study was obtained by parents and children from the age of 6. The study design followed the principles of the Declaration of Helsinki [23]. The study was approved by the local ethics committee and received ethical approval (EA2/238/19). All participating study centers have received ethical approval from their institutions.

2.3. Instruments

General and sociodemographic data such as diagnosis, study center, age, language spoken at home, and educational level from the age of 18 were collected. Variables such as “time since diagnosis” and “age of child at diagnosis” were calculated from diagnosis date and child’s age data.

Furthermore, self-constructed questions were asked about prior informedness, burden of diagnosis, and feelings of shame (Table 1) [20]. All questions were rated on a 5-point Likert scale, those on informedness

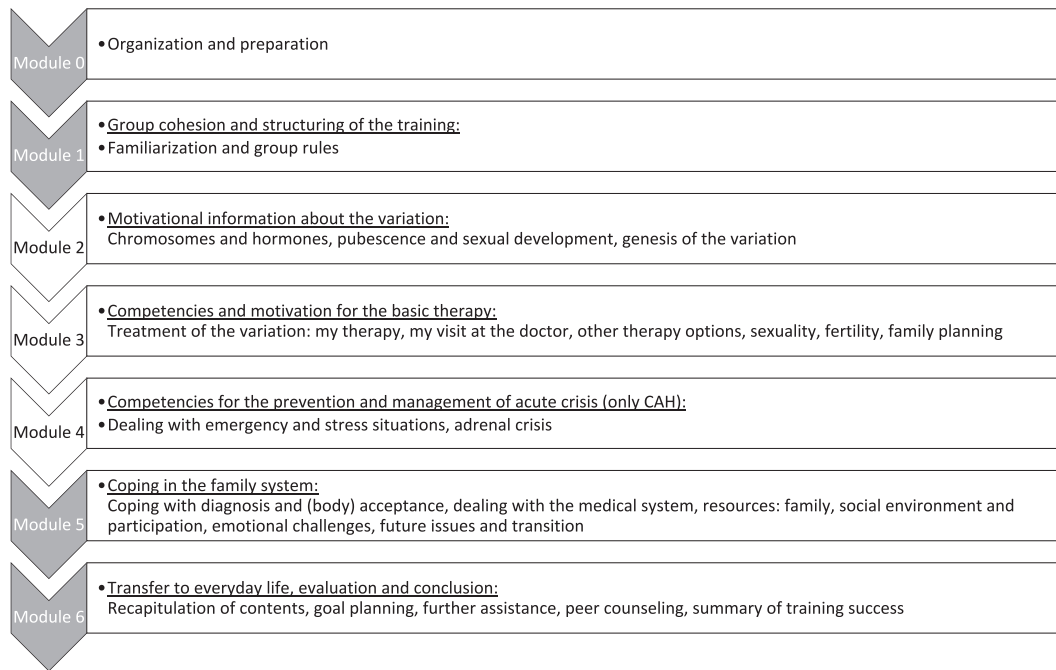


Fig. 1. Modules with the topics of the DSD curriculum.



Fig. 2. Diversity in course material: Alex* by Vielma©.

and feelings of shame included a “no response” option. The total score of the variable “feelings of shame” was obtained from the median of 5 questions on feelings of shame regarding the diagnosis and the body adapted of the dsd-LIFE study (Table 1) [24]. For the analysis, variables were dichotomized into high and low values (Table 1).

The concept of patient satisfaction in the context of patient education

is still poorly conceptualized and is based in this paper on a combination of concepts of patient satisfaction with health care services [25,26] and quality criteria of patient training [27]. The questionnaire to assess satisfaction with the training program was based on the ZUF-8 instrument, which was originally developed for assessing the satisfaction of hospitalized patients [26,28] and has already been used with an adapted

Table 1
Self-constructed variables with dichotomous categories.

Self-constructed variables	Dichotomous categories
How well do you feel informed about the diagnosis?	1 = well informed (1–3) 2 = less informed (4–6)
Have you ever been in contact to self-help groups for the diagnosis?	0 = no 1 = yes
How often do you think about your diagnosis? (Burden of diagnosis/thinking of diagnosis; participants with DSD)	1 = rarely (1/month, only for doctor's visits, never) 2 = often (each day, 1/week)
How strongly are you burdened by your child's diagnosis? Think about the last week. (parents)	1 = no or slight burden of diagnosis (0–1, not/little burdened) 2 = moderate to high burden of diagnosis (2–4)
Feelings of shame ((1) I am happy with the way I am. (2) My condition embarrasses me. (3) I have been teased and laughed at due to my condition. (4) I get embarrassed when I get changed in front of other people (during sport, on the beach). (5) My body embarrasses me.)	1 = high shame (1–3) 2 = low shame (4–6)

version to evaluate the satisfaction with ModuS trainings [19]. Several basic quality requirements for patient education programs were implemented in the items of the questionnaire by the study team [19,20,27,29].

For parents and young adults (18–24 years), 10 questions were used to assess general satisfaction, content quality, educators, and alignment of the training with their own expectations, needs, and management of the diagnosis (referred to as 'modified ZUF-8 adults'). After reversing the polarity, responses were rated on a 4-point Likert scale from 1 (=least favorable) to 4 (=most positive). The points were summed to a total score ranging from 10 (least satisfied) to 40 points (most satisfied). Two open questions with free-text answers provided the opportunity to offer feedback or suggestions for improvement of the program (supplement 1).

Children and adolescents' satisfaction was assessed using the 'modified ZUF-8 children' questionnaire with one item on general satisfaction rated on a 6-point scale related to German school grades, four items on the group process and the educators, among others, with a 5-point scale based on a smiley scale, and 2 open-ended questions (supplement 2). The analysis was conducted following a pole reversal, assigning 1 point for the lowest satisfaction and 5 respectively 6 points for the highest satisfaction. Here, the score ranged from a total of minimum 5 to a maximum of 26 points [26]. Subjects were included in the evaluations, if at least 80% of the items were answered.

A dichotomous satisfaction variable was created for analyzing influencing factors, categorizing as 'high satisfaction' (adults 30–40 points; children/adolescents 20–26 points) and 'low satisfaction' (adults 10–29 points; children/adolescents 5–19 points). Following Kriz et al. [30], the determination of a cut-off value for the ZUF instrument is based on the diagnosis. Thus, the published cut-off value of psychosomatic medicine was followed, which according to a receiver operating characteristic (ROC) analysis with Balance Error Scoring System (BESS) criterion is 24.5 and with Modelling Quality Indicator (MQI) criterion is 23.5 for a total score of 32 points [30]. Therefore, the adapted questionnaire sets off a cut-off value of 30 resulted proportionally for adults with a maximum score of 40 and a cut-off value of 20 for children/adolescents with a maximum score of 26.

2.4. Data collection

Before the training (baseline), the participants were asked questions about their socio-demographic and clinical background as well as their informedness, burden of diagnosis, and feelings of shame as part of the general evaluation. The evaluation of the participants' satisfaction with the training took place directly after the two-day training. Participants completed the online questionnaire via a tablet or their mobile phone

either directly whilst at the training site or at home afterwards. The access to the questionnaire was provided within 24 h after the training. Children were allowed to be assisted when filling it out by their parents or the training staff.

2.5. Data analysis

Descriptive statistics are presented for general and socio-demographic data as well as for participants' satisfaction (mean and standard deviation or frequencies with percentages), separately for children (ages 6–13), adolescents (ages 14–17), young adults (ages 18–24), and parents. The analysis included all subjects that consented to participate, attended at least one day of training, and entered the ZUF questionnaire. A difference of at least 10% of the total ZUF value was defined as a minimum clinically important difference (MCID). Missing data were not replaced. A Chi²-test was used to analyze the associations between the bivariate satisfaction variable and factors related to the diagnosis. All results were interpreted in an exploratory manner. Analyses were performed using SPSS, version 29.

The open questions were analyzed using thematic analysis based on Braun and Clark [31]. The answers were categorized deductively from the topics of the two open questions (positive feedback, suggestions for improvements) as well as inductively from the answers. The codes were listed according to their frequency and used for a deeper understanding of the quantitative data in the discussion and for further improvement of the curricula. The integration was driven by data and results.

3. Results

3.1. Participation

From 2020 to 2022, a total of 104 training sessions were conducted at the 5 study centers, including 18 training sessions for children, 37 for adolescents and young adults, and 49 for parents (Table 2). 102 children, 151 adolescents and young adults (95 adolescents 14–17 years, 56 young adults 18–24 years) and 380 parents participated in the training sessions, of which 139 were parents of newly diagnosed children (Table 3). The satisfaction assessment was completed by 89 (87.3%) children, 139 adolescents and young adults (92.1%), and 345 parents (90.8%).

3.2. Participant characteristics

The mean age (\pm standard deviation) was 10.6 ± 1.8 years in the group of children, 15.2 ± 1.0 years in the group of adolescents and 19.4 ± 1.8 years in the group of young adults. The mean age of the parents was 41.0 ± 9.3 years in the group with newly diagnosed children and 47.3 ± 7.8 years in the group of parents with children who had been diagnosed >2 years previously. Approximately 90% of the participating families spoke German at home (Table 4a, 4b). The level of education, based on the highest school-leaving qualification achieved, was reported as high by the majority of parents (66.1%) (Table 4b).

Table 2
Number of trainings undertaken by diagnosis and age group.

Diagnosis	Education group		
	Children (6–13 years)	Adolescents and young adults (14–24 years)	Parents of diagnosed children
Congenital adrenal hyperplasia	5	6	11
Klinefelter syndrome	2	12	13
Turner syndrome	7	8	9
Other DSD	4	11	16
Total	18	37	49

Table 3
Participation rate.

	Children (6–13 y.)			Adolescents and young adults (14–24 y.)			Parents of newly diagnosed children ¹			Parents of children with longstanding diagnosis ²		
	n	%	Drop-out	n	%	Drop-out	n	%	Drop-out	n	%	Drop-out
Informed about study	309	–	–	466	–	–	389	–	–	944	–	–
Consent to participate	121	100%	0.0%	178	100%	0.0%	170	100%	0.0%	292	100%	0.0%
Participation in education program	102	84.3%	15.7%	151	84.8%	15.2%	139	81.8%	18.2%	241	82.5%	17.5%
Questionnaire immediately after training	89	87.3%*	12.7%*	139	92.1%*	7.9%*	133	95.7%*	4.3%*	212	88.0%*	12.0%*

y. = years; n = number.

* Based on the population which participated in the education program.

¹ Diagnosis was made within the last 2 years.

² Including parents, primary caregivers and relatives;

Table 4a
Description of participating children, adolescents, and young adults.

	Children (6–13 y.)	Adolescents (14–17 y.)	Young adults (18–24 y.)
n	89	92	47
Age in years, mean ± SD	10.6 ± 1.8	15.2 ± 1.0	19.4 ± 1.8
Diagnosis			
Congenital adrenal hyperplasia, n (%)	29 (32.6)	15 (16.3)	4 (8.5)
Klinefelter syndrome, n (%)	17 (19.1)	42 (45.7)	16 (34.0)
Turner syndrome, n (%)	31 (34.8)	19 (20.7)	6 (12.8)
Other DSD, n (%)	12 (13.5)	16 (17.4)	21 (44.7)
Study center			
1, n (%)	36 (40.4)	30 (32.6)	23 (48.9)
2, n (%)	15 (16.9)	8 (8.7)	4 (8.5)
3, n (%)	18 (20.2)	8 (8.7)	4 (8.5)
4, n (%)	9 (10.1)	37 (40.2)	11 (23.4)
5, n (%)	11 (12.4)	9 (9.8)	5 (10.6)
Language spoken at home	n = 89	n = 92	n = 46
Only German, n (%)	68 (76.4)	74 (80.4)	32 (69.6)
German and other language, n (%)	9 (10.1)	9 (9.8)	6 (13.0)
Other language, n (%)	1 (1.1)	4 (4.3)	4 (8.7)
Not specified, n (%)	11 (12.4)	5 (5.4)	4 (8.7)

y. = years; n = number; SD = standard deviation.

3.3. Satisfaction assessment

Overall satisfaction was high in all groups. For children, the mean score was 24.4 ± 2.1 , and for adolescents, 23.5 ± 2.7 (Table 5). For adults 18 years and older, the mean satisfaction was at a similarly high level (young adults with DSD 36.0 ± 4.0 ; parents of newly diagnosed children 36.7 ± 3.2 ; other parents 36.6 ± 3.5) (Table 5 and 6). Regardless of diagnosis, participants' satisfaction was very high. Participants of TS training sessions consistently showed the highest scores (Tables 5, 6). However, differences shown were not clinically relevant.

The language spoken at home was not relevantly associated with satisfaction across the groups. Except for adolescents, all groups showed a slightly lower satisfaction score on average if a language other than German was spoken at home (Tables 5, 6). Satisfaction by parental educational level did not differ considerably among parents with a low, medium, and high level (35.8 ± 4.4 , 37.4 ± 2.5 , and 36.5 ± 3.5 respectively) (Table 6).

The time since diagnosis did not make a difference to the satisfaction of the parents on average. Parents of newly diagnosed children scored 36.7 ± 3.2 compared to parents who already have known the diagnosis for >10 years (38.0 ± 2.1). The age of the child at diagnosis was also not associated with the satisfaction of the parents (Table 6). For children, adolescents, and young adults, the frequency of thinking about the

diagnosis was not associated with the level of satisfaction (Table 7a). Regarding feelings of shame, both participants with low and those with high feelings of shame showed high satisfaction (Table 7a). The level of information about the diagnosis was not related to satisfaction in adolescents and young adults. Only for children a tendency was found that prior knowledge was associated with lower satisfaction. Even if the participants were in contact with the self-help group before the training, the satisfaction of all participants was high (Table 7a).

Around half of the parents felt a great burden due to the diagnosis of their child. Regardless of the extent of this burden, parents were very satisfied with the training (Table 7b). Although many parents felt well informed prior to the training, they were very satisfied regardless of whether their child had been newly diagnosed or for a longer time (79.1% and 91.0%, respectively). Parents who had no previous contact with self-help groups or felt less informed also showed high satisfaction (Table 7b).

3.4. Qualitative findings on influencing factors on satisfaction

Questioning young adults, parents as well as children and adolescents about what they like most revealed that the greatest value of the training was the exchange of information and experiences (Table 8).

“The feeling of no longer being alone. Having our questions answered and being able to talk openly about our concerns about the diagnosis” (mother, 45–241).

For persons with DSD, the trainings offered the opportunity to stabilize their own self-esteem and to realize that one is not alone which might otherwise be the impression they get in regular consultation hours.

“I thought the structure of the training was great, the topics of conversation and the exchange with each other. The feeling of not being alone and, for example, being able to admit grief is a pleasant feeling. The training made me feel special and anything but worthless” (young adult, 22 years, 45–108).

The majority emphasized the comfortable and open atmosphere of the groups. Medical and psychosocial content, educator skills, and methods supported their subjective perception of high satisfaction.

“I was well and honestly informed about the topic all round. The educators always hit the right nerve and were also able to deal well with the emotions of the participants. It was very helpful to be able to talk to people who are also affected and to do so with scientific support. In particular, this helped to immediately relativize or correct false information that may be spread by other means” (father, 44–135).

The trainings were said to provide an extension of regular care, as there is more time for questions, practicing skills (e.g. injection training for CAH), developing resilience and empowerment strategies and benefiting from the experiences of others. Although many families already felt well informed prior to the trainings, they rated the added value of the training in the intensive discussion of topics and the supervised exchange and contact with other people with the same condition.

Table 4b
Description of parents of children with new and with longstanding diagnoses.

	Parents of newly diagnosed children ¹	Parents of children with longstanding diagnosis ²	Total
n (number of parents)	133	212	345
Age in years, mean ± SD (number included)	41.0 ± 9.3 (n = 132)	47.3 ± 7.8 (n = 208)	44.8 ± 8.9 (n = 340)
Study center			
1, n (%)	32 (24.1)	73 (34.4)	105 (30.4)
2, n (%)	29 (21.8)	30 (14.2)	59 (17.1)
3, n (%)	5 (3.8)	36 (17.0)	42 (11.9)
4, n (%)	26 (19.5)	59 (27.8)	85 (24.6)
5, n (%)	41 (30.8)	14 (6.6)	55 (15.9)
Educational level			
Low, n (%)	n = 120 16 (13.3)	n = 191 11 (5.8)	n = 311 27 (8.7)
Medium, n (%)	32 (26.7)	49 (25.7)	81 (26.0)
High, n (%)	72 (60.0)	131 (68.6)	203 (65.3)
Language spoken at home			
Only German, n (%)	n = 132 113 (85.6)	n = 208 179 (84.4)	n = 340 292 (85.9)
German and other language, n (%)	12 (9.1)	19 (9.0)	31 (9.1)
No German, n (%)	5 (3.8)	7 (3.3)	12 (3.5)
Not specified, n (%)	2 (1.5)	3 (1.4)	5 (1.5)
Diagnosis of child			
Congenital adrenal hyperplasia, n (%)	38 (28.6)	44 (20.8)	82 (23.8)
Klinefelter syndrome, n (%)	29 (21.8)	75 (35.4)	104 (30.1)
Turner syndrome, n (%)	20 (15.0)	56 (26.4)	76 (22.0)
Other DSD, n (%)	46 (34.6)	37 (17.5)	83 (24.1)
Time since diagnosis			
lt; 2 years (newly diagnosed), n (%)	n = 133 133 (100.0)	n = 105 –	n = 238 133 (55.9)
2–10 years, n (%)	–	74 (70.5)	74 (31.1)
gt; 10 years, n (%)	–	31 (29.5)	31 (13.0)
Age of child at diagnosis			
Before birth - 2 years, n (%)	n = 101 49 (48.5)	n = 105 62 (59.0)	n = 206 111 (53.9)
2–10 years, n (%)	13 (12.9)	26 (24.8)	39 (18.9)
11–16 years, n (%)	32 (31.7)	11 (10.5)	43 (20.9)
gt; 16 years, n (%)	7 (6.9)	6 (5.7)	13 (6.3)
Burden of diagnosis			
No or slight burden of diagnosis, n (%)	n = 109 49 (45.0)	n = 182 95 (52.2)	n = 291 144 (49.5)
Moderate to high burden of diagnosis, n (%)	60 (55.0)	87 (47.8)	147 (50.5)

n = number; SD = standard deviation.

¹ Diagnosed within the last 2 years.² Including parents, primary caregivers and relatives;

“Although I already felt well informed beforehand, I learned a lot of new things as a result of the intensive training” (mother, 45–205).

“The opportunity to exchange more intensively with everyone involved in our care. The consultation hours are also good, but the two intensive training days gave us the chance to focus more on the topic” (mother of a newly

Table 5
Satisfaction with education program, separately for children, adolescents, and young adults.

	Modified ZUF-8 children ¹ (Mean ± SD)		Modified ZUF-8 adults ² (Mean ± SD)
	Children (6–13 y.)	Adolescents (14–17 y.)	Young adults (18–24 y.)
n ³	87	91	47
Overall	24.4 ± 2.1	23.5 ± 2.7	36.0 ± 4.0
Diagnosis			
Congenital adrenal hyperplasia	24.6 ± 1.7 (n = 28)	22.7 ± 2.4 (n = 15)	37.0 ± 2.9 (n = 4)
Klinefelter syndrome	22.8 ± 2.8 (n = 17)	23.1 ± 3.0 (n = 42)	35.0 ± 4.9 (n = 16)
Turner syndrome	24.8 ± 1.9 (n = 30)	24.7 ± 2.2 (n = 18)	35.5 ± 4.4 (n = 6)
Other DSD	24.8 ± 1.4 (n = 12)	23.8 ± 2.5 (n = 16)	36.8 ± 3.4 (n = 21)
Language spoken at home			
Only German	24.5 ± 1.8 (n = 66)	23.3 ± 2.9 (n = 73)	36.8 ± 3.2 (n = 32)
German and/or other language	23.7 ± 2.9 (n = 21)	24.2 ± 1.6 (n = 18)	34.1 ± 5.3 (n = 14)

y. = years; n = number; SD = standard deviation.

¹ Total score ranging from 5 to 26. High scores correspond to high satisfaction.² Total score ranging from 10 to 40. High scores correspond to high satisfaction.³ At least 80% of the items had to be answered.

diagnosed child, 45–2).

“I understood my diagnosis for the first time” (child, 10 years, 45–142).

The most common suggestions for improvement were related to group composition, time management of training, and content, e.g. missing topics. Many expressed the need for future training and follow-up meetings, as well as networking among participants. More orientation towards individual topics during the training and the opportunity for individual medical and/or psychological counseling were frequently mentioned (Table 8).

4. Discussion and conclusion

4.1. Discussion

The present study revealed a high level of overall satisfaction with the new educational program for young people with DSD and their parents in all groups. High satisfaction with training was independent of burden of diagnosis, feelings of shame, or prior informedness about the diagnosis.

Using the widely accepted concept for patient education in Germany – ModuS – the approved modular structure and practical experience from implementation and realization were used; moreover, the results of evaluations could be compared [17,19,32]. Overall, a similarly high level of satisfaction with the training program could be achieved as for other ModuS programs [17,19]. Satisfied patients have a better understanding of and higher adherence to the therapy, attend regular check-ups, feel more responsible for their health and have fewer health problems [25,33].

To address the diverse spectrum of DSD diagnoses, four tailored curricula were created in collaboration with the respective self-help groups. Avoiding terms like *disease* or *disorder* the curricula embraced a universal, gender-sensitive language, emphasizing *variation* or *particularity* instead. The results showed no clinically relevant differences in participant satisfaction, indicating that the education program was tailored for the diagnostic groups with its unique openness and emphasis on gender diversity. It underlines the central role of well-trained

Table 6
Satisfaction with education program, separately for parents of children with new and longstanding diagnoses.

	Modified ZUF-8 adults ² (Mean ± SD)		Total
	Parents of newly diagnosed children ¹	Parents of children with longstanding diagnosis ³	
n ⁴	132	209	341
Overall	36.7 ± 3.2	36.6 ± 3.5	36.6 ± 3.4
Diagnosis of child			
Congenital adrenal hyperplasia	37.0 ± 3.2 (n = 37)	37.1 ± 2.7 (n = 43)	37.1 ± 2.9 (n = 80)
Klinefelter syndrome	36.2 ± 3.1 (n = 29)	35.4 ± 4.2 (n = 75)	35.6 ± 3.9 (n = 104)
Turner syndrome	37.1 ± 2.2 (n = 20)	37.1 ± 2.7 (n = 54)	37.1 ± 2.5 (n = 74)
Other DSD	36.5 ± 3.7 (n = 46)	37.6 ± 3.2 (n = 37)	37.0 ± 3.5 (n = 83)
Language spoken at home			
Only German	n = 131 36.8 ± 2.9 (n = 112)	n = 205 36.8 ± 3.4 (n = 176)	n = 336 36.8 ± 3.2 (n = 288)
German and/or other language	35.4 ± 4.6 (n = 19)	35.6 ± 3.6 (n = 29)	35.5 ± 4.0 (n = 48)
Educational level			
Low	n = 119 35.9 ± 4.4 (n = 15)	n = 188 35.7 ± 4.4 (n = 10)	n = 307 35.8 ± 4.3 (n = 25)
Medium	37.6 ± 2.3 (n = 32)	37.2 ± 2.7 (n = 47)	37.4 ± 2.5 (n = 79)
High	36.2 ± 3.2 (n = 72)	36.7 ± 3.6 (n = 131)	36.5 ± 3.5 (n = 203)
Time since diagnosis			
lt; 2 years	–	–	n = 236 36.7 ± 3.2 (n = 132)
2–10 years	–	–	36.6 ± 3.8 (n = 73)
gt; 10 years	–	–	38.0 ± 2.1 (n = 31)
Age of child at diagnosis			
Before birth - 2 years	n = 100 36.2 ± 3.2 (n = 48)	n = 104 37.5 ± 2.8 (n = 62)	n = 204 36.9 ± 3.0 (n = 106)
2–10 years	37.7 ± 2.8 (n = 13)	36.5 ± 4.4 (n = 25)	36.9 ± 3.9 (n = 36)
11–16 years	36.6 ± 3.6 (n = 32)	35.0 ± 4.8 (n = 11)	36.2 ± 3.9 (n = 40)
gt; 16 years	36.1 ± 4.1 (n = 7)	38.0 ± 1.5 (n = 6)	37.0 ± 3.2 (n = 12)

n = number; SD = standard deviation.

¹ Diagnosed within the last 2 years.

² Total score ranging from 10 to 40. High scores correspond to high satisfaction.

³ Including parents, primary caregivers and relatives.

⁴ At least 80% of the items had to be answered.

educators in providing an effective program and ensuring participant satisfaction through their attitude and language [19].

A majority of the participants in this study felt well informed before the training. This might be explained as a result of many of the families were treated in one of the participating DSD centers in Germany. The present study found no differences in the extent of prior knowledge and satisfaction with the training. It can be expected that existing prior knowledge and intellectual engagement with diagnosis-related issues, thus a high level of health literacy, could lead to better understanding of the content and thus to a high level of satisfaction. Therefore, the integration of existing knowledge with new knowledge as well as the integration of new skills for individual empowerment are then experienced successfully [30]. For people with low prior knowledge, any increase in knowledge could lead to high satisfaction. Therefore, the individual requirements of the participants should always be considered, and groups should be formed as homogeneously as possible in order to maximize the increase in knowledge. Nevertheless, mixed groups, e.g., with parents of newly diagnosed children and parents of children diagnosed years ago, could also lead to fruitful discussions.

According to current recommendations for the care of people with DSD, these target groups need improved psychosocial care [7,15]. Families are often scared of the impact of the diagnosis on the child's health. They are worried about stigmatization and disadvantages for their child and think about the possible effects of the diagnosis on the development of the reproductive organs, fertility, and gender identity. These are topics that are often associated with shame and taboos, and many people with DSD are concerned about physical changes, and visions for their future. Nevertheless, satisfaction was high in all groups, regardless of burden of diagnosis and feelings of shame. This reflects that the psychosocial counseling and the open atmosphere in the training sessions addressed these fears quite well and the participants benefited from the exchange with their peers, which was the most frequent positive feedback given in the free-text responses. In general, social contact, informal exchange and psychosocial content are at least as important as medical content. At the same time, this is also a limitation of patient education. It cannot address all individual questions in depth but can only touch on example issues and facilitate the development of solution strategies. As an answer to the open questions, the need for more individual psychosocial counseling was often expressed. Thus, the boundaries of group training and counseling compared to therapy become apparent. Group training can give the feeling that no one is "alone" with the diagnosis. It can help to provide contacts, either to peers or professionals, and thus contribute to the improvement of health in the long term and avoid psychosocial secondary costs [34]. However, patient training cannot replace individual therapy, but group training should be an additional element in the multidisciplinary care of people with DSD and their families whether they are treated in specialized DSD centers or not.

4.2. Strengths and limitations

This was the first evaluation of the satisfaction with a newly developed modular, age- and diagnosis-specific education program for young people with DSD and their parents in Germany. Further strengths included a broad spectrum of DSD diagnoses, the participation of a number of specialized hospitals across Germany and of various self-help groups in the development and evaluation process.

However, several limitations have to be noted. First, a general limitation of the ZUF-8 instrument used, is the skewed distribution of the responses resulting in a high average satisfaction and low variance overall [30]. Binary logistic regression was used to identify predictors of satisfaction, but no interpretable results could be obtained due to the low variance. Therefore, only descriptive data were presented. Second, a

Table 7a

Associations between diagnosis-related characteristics and satisfaction with the education program, separately for children, adolescents and young adults.

	Children (6–13 y.)			Adolescents (14–17 y.)			Young adults (18–24 y.)		
	Low satisfaction (score 5–19)	High satisfaction (score 20–26)	p-value	Low satisfaction (score 5–19)	High satisfaction (score 20–26)	p-value	Low satisfaction (score 10–29)	High satisfaction (score 30–40)	p-value
Burden of diagnosis (thinking of diagnosis)	n = 84 (100%)			n = 87 (100%)			n = 44 (100%)		
Rarely, n (%) ¹	3 (3.6)	42 (50.0)	0.101	6 (6.9)	40 (46.0)	0.599	1 (2.3)	18 (40.9)	0.842
Often, n (%) ¹	0 (0.0)	39 (46.4)		7 (8.0)	34 (39.1)		1 (2.3)	24 (54.5)	
Feeling informed about diagnosis	n = 75 (100%)			n = 86 (100%)			n = 46 (100%)		
Less informed, n (%) ¹	1 (1.3)	8 (10.7)	0.006	2 (2.3)	9 (10.5)	0.761	0 (0.0)	4 (8.7)	0.655
Well informed, n (%) ¹	0 (0.0)	66 (88.0)		11 (12.8)	64 (74.4)		2 (4.3)	40 (87.0)	
Contact to self-help group	n = 73 (100%)			n = 81 (100%)			n = 46 (100%)		
Yes, n (%) ¹	1 (1.4)	13 (17.8)	0.039	3 (3.7)	7 (8.6)	0.105	0 (0.0)	7 (15.2)	0.540
No, n (%) ¹	0 (0)	59 (80.8)		8 (9.9)	63 (77.8)		2 (4.3)	37 (80.4)	
Feelings of shame	n = 73 (100%)			n = 84 (100%)			n = 44 (100%)		
Low shame, n (%) ¹	2 (2.7)	56 (76.7)	0.466	10 (11.9)	55 (65.5)	0.250	1 (2.3)	32 (72.7)	0.403
High shame, n (%) ¹	0 (0)	15 (20.5)		1 (1.2)	18 (21.4)		1 (2.3)	10 (22.7)	

y. = years; n = number.

¹ Percentage of all.

Table 7b

Associations between diagnosis-related characteristics and satisfaction with the education program, separately for parents of children with new and with longstanding diagnoses.

	Parents with newly diagnosed children ¹			Parents of children with longstanding diagnosis ²			Total		
	Low satisfaction (score 10–29)	High satisfaction (score 30–40)	p-value	Low satisfaction (score 10–29)	High satisfaction (score 30–40)	p-value	Low satisfaction (score 10–29)	High satisfaction (score 30–40)	p-value
Burden of diagnosis	n = 108 (100%)			n = 180 (100%)			n = 288 (100%)		
No or slight burden of diagnosis, n (%) ³	1 (0.9)	48 (44.4)	0.243	2 (1.1)	92 (51.1)	0.346	3 (1.9)	140 (48.6)	0.130
Moderate to high burden of diagnosis, n (%) ³	4 (3.7)	55 (50.9)		4 (2.2)	82 (45.6)		8 (2.8)	137 (47.6)	
Feeling informed about diagnosis	n = 129 (100%)			n = 200 (100%)			n = 329 (100%)		
Less informed, n (%) ³	1 (0.8)	21 (16.3)	0.979	0 (0.0)	11 (5.5)	0.516	1 (0.3)	32 (9.7)	0.775
Well informed, n (%) ³	5 (3.9)	102 (79.1)		7 (3.5)	182 (91.0)		12 (3.6)	284 (86.3)	
Contact to self-help group	n = 124 (100%)			n = 200 (100%)			n = 324 (100%)		
Yes, n (%) ³	2 (1.6)	26 (21.0)	0.342	5 (2.5)	52 (26.0)	0.010	7 (2.2)	78 (24.1)	0.010
No, n (%) ³	3 (2.4)	93 (75.0)		2 (1.0)	141 (70.5)		5 (1.5)	234 (72.2)	

n = number.

¹ Diagnosed within the last 2 years.

² Including parents, primary caregivers and relatives.

³ Percentage of all.

possible bias may have occurred due to the selection of participants, who were particularly highly motivated, since the training was offered for the first-time and within a context of a research project. The responses may have also been affected by social desirability or the motivation of long-term patients to support the study and ensure the success of the program. Third, the educational program provided was only in German, so not all potential population groups could be addressed. Fourth, the recruited sample consisted predominantly of highly educated participants of German speaking households who already felt well informed prior to the training. Therefore, the results of this survey cannot be generalized, as education level and language skills in particular contribute to satisfaction with training interventions [35] and are crucial factors for understanding health-related learning content [36]. Fifth, the children were allowed to answer the questionnaire with the help of their parents. It is unclear whether this had any influence on the responses. Sixth, a major issue in the research of learning settings is that it is difficult to standardize the situations. With a structured curriculum and the qualification of educators according to standardized specifications by ModuS [29], the training was standardized as much as possible. However, the

individual personalities of the educators, the dynamics of the group, the conditions at the study centers (e.g. COVID-19 restrictions), and other individual factors may have led to bias. Participants may have experienced emotions like sadness, anger, or despair, or may have become more aware of the impact of their diagnosis, leading to new questions and a need for more time than 24 h to reflect on training satisfaction. Seventh, although the barriers to participation were kept as low as possible (e.g. no fees), it remains unclear why many eligible persons with DSD did not want to participate. Finally, it remains unclear whether the results were influenced by non-responders if they may have been less satisfied than those who participated in the evaluation.

4.3. Innovation

The research presented here outlines the development of the first educational program for children, adolescents, and young adults with DSD and their families, aimed at providing comprehensive health information and empowerment. This interdisciplinary, modular program serves as a foundation for informed decision-making, even for minors,

Table 8

Frequencies of codes from qualitative analysis of the answers of the open questions of the modified ZUF-8.

Parents ¹ and young adults ² (Total number of codes = 573)		Children and adolescents ³ (Total number of codes = 181)	
Positive feedback			
	Counts		Counts
exchange	163	exchange	47
group atmosphere and openness	93	methods	32
content (medical and psychosocial)	83	increase in knowledge	27
trainer skills	53	group atmosphere and openness	25
methods	34	medical content	21
team and support	25	trainer skills	13
room	15	support	5
own positive feelings	14	food supply	4
time management	12	psychosocial content	4
increase in knowledge	12	peer consultation	2
peer consultation	10	medical consultation	1
organization	8		
medical and psychosocial consultation	8		
individual speaking	7		
food supply	6		
Suggestions for improvements			
time management	62	nothing/everything fine	92
content	46	group composition	20
nothing/ everything fine	53	methods	18
group composition	30	time management	13
organization	29	organization	6
methods	25	food supply	5
future ideas	22	content	5
follow-up meeting and networking	17	team	1
peer consultation	13	peer meeting	1
food and room supply	12	remaining questions	1
individual consultation	11		
survey questions	6		
group management	3		
trainer	2		
remaining questions	2		

¹ Including parents, primary caregivers and relatives.

² Age of 18–24 years.

³ Age of 6–17 years.

aligning with current standards for DSD treatment. The inclusion of self-help groups throughout the project facilitated the adaptation of materials and concepts to the needs of the target group. The program emphasizes gender-neutral, non-judgmental language and avoids terms like *disease* or *disorder*. Adapting a well-known patient education concept for chronic diseases (ModuS) to this philosophy posed a challenge. The result is an innovative approach focusing more on acceptance as a biological feature of sexual development rather than perception as a disease. It encourages a discussion about “classical norms” and is intended to liberate from the social pressure to fit in. The significant value of the program is not only in increasing knowledge and skills, but also in its emphasis on shifting attitudes towards acceptance of the body and sex/gender, which could thus reduce the need for psychosocial support. The high level of participant satisfaction reflects the appropriate approach for this target group. High satisfaction with health services can lead to improved use of health care services, informed and confident medical decision making, and improved health [25]. Improved self-acceptance and medical decision-making skills may lead to higher adherence and an improved transition of adolescents to adult health care, thus potentially leading to reduced health care costs. In order to investigate this, it is necessary to continue the training sessions and to conduct a long-term follow-up evaluation. A subsequent project might involve creating digital educational modules integrated with in-person psychoeducational components.

4.4. Conclusion

The developed education program is - to the best of the authors' knowledge - the first formally standardized and institutionalized program worldwide for young people with DSD. The satisfaction levels with the new education program including tailored diagnosis-specific medical information, peer consultation, and psychological support were high among varying groups of persons with DSD and their parents. Further analyses need to determine long-term effects of the program in terms of wellbeing and increased knowledge. The program can support people with DSD and their parents in coping with the diagnosis and should be an integral part of multidisciplinary care. Additional research is required to develop a conceptual framework for assessing patient satisfaction regarding educational interventions and to explore its impact on health outcomes and the use of health care.

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CRedit authorship contribution statement

Sabine Wiegmann: Writing – review & editing, Writing – original draft, Visualization, Project administration, Methodology, Formal analysis. **Ralph Schilling:** Writing – review & editing, Validation, Methodology, Formal analysis. **Mirja Winter:** Writing – review & editing, Investigation. **Martina Ernst:** Writing – review & editing, Investigation. **Katja Wechsung:** Writing – review & editing, Investigation. **Ute Kalender:** Writing – review & editing, Investigation. **Barbara Stöckigt:** Writing – review & editing, Methodology, Investigation. **Annette Richter-Unruh:** Writing – review & editing, Investigation, Conceptualization. **Olaf Hiort:** Writing – review & editing, Investigation, Conceptualization. **Ulla Döhnert:** Writing – review & editing, Investigation. **Louise Marshall:** Writing – review & editing, Investigation. **Julia Rohayem:** Writing – review & editing, Investigation, Conceptualization. **Klaus-Peter Liesenkötter:** Writing – review & editing, Investigation. **Martin Wabitsch:** Writing – review & editing, Conceptualization. **Gloria Herrmann:** Writing – review & editing, Investigation. **Gundula Ernst:** Writing – review & editing, Conceptualization. **Stephanie Roll:** Writing – review & editing, Validation, Supervision, Methodology, Formal analysis, Conceptualization. **Thomas Keil:** Writing – review & editing, Supervision, Methodology, Funding acquisition, Data curation, Conceptualization. **Uta Neumann:** Writing – review & editing, Supervision, Project administration, Methodology, Funding acquisition, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

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