Shaping Parents: Impact of Contrasting Professional Counseling on Parents’ Decision Making for Children with Disorders of Sex Development

Jürg C. Streuli, MD,* Effy Vayena, PhD,* Yvonne Cavicchia-Balmer, BA,* and Johannes Huber, MD, PhD†

*Institute of Biomedical Ethics, University of Zürich, Zürich, Switzerland; †Department of Urology, University Hospital Carl Gustav Carus, Technical University of Dresden, Dresden, Germany

DOI: 10.1111/jsm.12214

ABSTRACT

Introduction. The management of disorders or differences of sex development (DSD) remains complex, especially with respect to parents' decision for or against early genitoplasty. Most parents still tend to disfavor postponing surgery until the child is old enough to provide consent.

Aim. To identify the determinants of parental decisions for or against early sex assignment surgery in DSD children, and in particular to assess the influence of contrasting behavior of health-care professionals and the information they dispense.

Methods. Preliminary data analysis from a focus group identified two broad approaches to counseling information. Two six-minute counseling videos were produced on this basis: one medicalized, by an endocrinologist, the other demedicalized, by a psychologist. Third-year medical students (N = 89) were randomized to watch either video as prospective parents and report its impact on their decision in a self-administered questionnaire.

Main Outcome Measures. Statistical analysis of questionnaire responses regarding decisions for or against surgery, including self-assessed impact of potential determinants.

Results. Thirty-eight of eighty-nine “parents” (43%) chose early surgery for “their” child, including 27/41 “parents” (66%) shown the medicalized video vs. 11/48 (23%) shown the demedicalized video (P < 0.001). Desired aims for “their” child also differed significantly depending on the counseling approach viewed. Yet “parents” perceived their personal attitudes on a four-point Likert scale as the main influence on their decision although their “attitude” was significantly shaped by the video.

Conclusions. Parental decisions concerning early sex assignment surgery for DSD children depend on the health professional counseling received, to a degree of which neither parents nor professionals appear fully aware. In the absence of conclusive data for or against early surgery, there is a danger of medicalized or demedicalized parentalism resulting in irreversible and inadequately grounded decisions, regardless of the consensus statement of 2005 and the subsequent call for multidisciplinary management. Streuli JC, Vayena E, Cavicchia-Balmer Y, and Huber J. Shaping parents: Impact of contrasting professional counseling on parents’ decision making for children with disorders of sex development. J Sex Med **,**,**,**,**, **.

Key Words. Disorders of Sex Development; Informed Consent; Intersex; Normality; Pediatric Endocrinology; Pediatric Psychology

Introduction

The management of disorders or differences of sex development (DSD), also known as intersex, remains complex, with special respect to the parental decision as to genitoplasty in early
childhood. In most ethical and professional guidelines and recommendations, informed parental consent is not simply a legal requirement but is also considered important for long-term outcome in terms of satisfaction, adherence, and resilience [1–6].

Parents consider it their responsibility or are considered to be responsible for deciding for or against a particular treatment regime, including genital surgery [1,2,7]. However, as shown by Crawford et al., parents might see surgery as obvious and necessary, without experiencing it as something that involved a decision-making process [8]. Despite lack of evidence concerning outcome data and despite being informed about issues such as their child’s potential loss of sexual sensation, parents tend to disfavor postponing surgery until the child is old enough to provide its own consent [3,4,7–11].

The origin of this readiness to authorize childhood surgery is unclear. Some authors invoke parental perception of social pressure or parents’ long-held attitudes (e.g., sexual norms) [11]. Other see parents as influenced primarily by the behavior of health-care professionals and the information they dispense, which can appear medicalized or demedicalized, mainly depending on the context, cause, and proposed solution [7–13]. These nuances are reflected in the nomenclature, with some authors preferring the term disorder of sex development, denoting a defined “medical condition,” covered by insurance and others, whereas others refer to difference of sex development, as less stigmatizing and demeaning, and more sensitive in that it denotes an individual rather than a clinical condition [12–14].

Parents may act and decide differently depending on whether their primary information source is essentially medical, speaking of “disorders,” “congenital malformation,” and “surgical options,” or whether it consists of less pathologizing and more supportive information combined with coping strategy counseling as described, for instance, by the DSD Consortium [14,15]. Although current recommendations stress the importance of multilateral approaches including experts with different perspectives, professionals’ personal opinions on what would be preferable, bearing in mind their evaluation of the family and child, and their wider social and cultural context, may remain pivotal [15–17].

Aims

Research on the effect of differences in counseling information content and mode of delivery is virtually nonexistent. Our aim was to investigate the impact of information and behavior within multidisciplinary groups on parental decision making in the DSD setting.

Methods

The study was conducted at Zurich University Hospital, Switzerland, in 2011–2012, at the Institute of Biomedical Ethics at the University of Zurich liaising with the School of Applied Psychology at the Zurich University of Applied Sciences [18]. No patient was directly involved, but informed consent was obtained from participating volunteers, and the study was approved by the institutional review board.

Focus Group Data Collection

We set up a focus group comprising one parent of a DSD child, two (now adult) DSD activists (of whom one was a former DSD patient), and members of a hospital-based DSD team (pediatric endocrinologist, plastic/urogenital surgeon, neonatologist, psychologist, and medical ethicist [J.S.]). The 2-hour discussion, led by J.S., focused on counseling and educating the parents of a child born with ambiguous genitalia/DSD. The session was video’d, transcribed verbatim, analyzed using qualitative data analysis software (ATLAS/ti. 2011, Version 6.2., Scientific Software Development, Berlin, Germany), and authenticated by participant review [19,20].

Video Information Talk

Based on citations and concepts emerging from the focus group data we wrote two different scripts for an introductory patient information talk. One was written as if presented by an endocrinologist, the other as if presented by a psychologist. Each script was reviewed by the endocrinologist and the psychologist, and each certified “their” version as accurate. Based on the scripts we produced two six-minute videos in which J.S. acted as endocrinologist and psychologist, respectively (see online content for the English-language video transcripts).

Recruitment of Parental Role-Players and Video Data Collection

We recruited parental role-players from among third-year medical students starting their course in clinical ethics. Groups of 10 to 12 students were randomized to view the medicalized talk by an
endocrinologist (n = 41) or the demedicalized talk by a psychologist (n = 48).

All parental role-players were first shown a video asking them to imagine that they had just become parents of a child with ambiguous genitalia, whose (future) gender midwives and doctors were unable to identify, and that the second video they were about to watch was their first contact with a specialist able to counsel and inform them about their child’s condition. The students were given no opportunity to discuss the impact of this second video with other groups nor were they informed about the existence of an alternative second video.

**Main Outcome Measures**

After students had viewed the video, they had 5 minutes in which to answer a self-completed questionnaire assessing their understanding of DSD and asking them to decide for or against surgery for “their” child. They were also asked to rate their perceived security on a 10-point Likert scale (1 = not secure at all, 10 = absolutely secure) and 16 predefined potential determinants of their treatment decision on a four-point Likert scale.

**Data Extraction and Statistical Analysis**

Categorical data are presented as absolute and relative frequencies, and continuous data as means and standard deviations. Range is provided for selected values. We used the chi-square test and Student’s t-test for exploratory univariate statistics. A significance level of \( P < 0.05 \) was used in all tests. We performed all calculations using IBM SPSS statistics software (version 19; IBM Corp., Armonk, NY, USA).

**Results**

**Focus Group Data Analysis**

Participants’ views revealed two basic perspectives on the issue resulting in two broadly different approaches to informing, counseling, and supporting parents and their affected children. The medical professionals tended to medicalize, defining the child and its condition or behavior as a medical problem or illness that mandated or licensed the medical professional to offer a specific treatment [13]. The parent, patient, activist, and psychologist, on the other hand, tended to demedicalize the issue by stressing the importance of the child’s social world, seeing the main task as offering the child professional support in its environment (Table 1).

**Table 1** The two approaches emerging from the focus group discussion of DSD

<table>
<thead>
<tr>
<th>Medicalized approach</th>
<th>Demedicalized approach</th>
</tr>
</thead>
<tbody>
<tr>
<td>DSD is a static disorder with an inherent psychosocial component.</td>
<td>DSD is a dynamic disorder characterized by context-dependent impairment.</td>
</tr>
<tr>
<td>DSD and DSD-like conditions are disorders requiring treatment.</td>
<td>Development, resilience, and coping strategies need to be fostered.</td>
</tr>
<tr>
<td>Treatment should be function oriented.</td>
<td>Treatment should be geared to interests and capabilities.</td>
</tr>
<tr>
<td>Functions are biologically determined.</td>
<td>Functions are subject to multifactorial influences.</td>
</tr>
<tr>
<td>Treatment regimes are predetermined.</td>
<td>Support is individualized, adaptable, and dynamic.</td>
</tr>
<tr>
<td>The child is a passive agent in defining the problem.</td>
<td>The child and his or her family are active agents in defining the problem.</td>
</tr>
<tr>
<td>Aid should be hospital based.</td>
<td>Aid should be community based.</td>
</tr>
</tbody>
</table>

**Table 2** Characteristics of the study groups exposed to the two types of video information

<table>
<thead>
<tr>
<th>Variable</th>
<th>Population (N = 89)</th>
<th>Medicalized video (n = 41)</th>
<th>Demedicalized video (n = 48)</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, year (mean ± SD)</td>
<td>23.6 ± 3.3</td>
<td>23.5 ± 3.0</td>
<td>23.6 ± 3.6</td>
<td>0.95</td>
</tr>
<tr>
<td>Female sex, n (%)</td>
<td>50 (56)</td>
<td>24 (59)</td>
<td>28 (54)</td>
<td>0.68</td>
</tr>
<tr>
<td>Prior knowledge of DSD, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>None</td>
<td>6 (7)</td>
<td>4 (10)</td>
<td>2 (4)</td>
<td>0.30</td>
</tr>
<tr>
<td>Little</td>
<td>33 (37)</td>
<td>16 (39)</td>
<td>17 (35)</td>
<td>0.73</td>
</tr>
<tr>
<td>More than in the video</td>
<td>38 (43)</td>
<td>16 (39)</td>
<td>22 (46)</td>
<td>0.52</td>
</tr>
<tr>
<td>Aware of difference between intersex and transsex</td>
<td>56 (63)</td>
<td>24 (59)</td>
<td>32 (67)</td>
<td>0.43</td>
</tr>
</tbody>
</table>

**Characteristics of Student Role-Player Participants**

Of 94 potential participants, four did not give consent and one returned a blank questionnaire. Of the included 89 students, 48 (54%) were exposed to the “medicalized” video, and 41 (46%) to the “demedicalized” video (Figure 1).

Participants’ characteristics showed comparable gender, age, and self-assessed prior knowledge between video groups (Table 2). Mean age was
23.6 ± 3.3 years (range 21–39 years). Male participants were older (27.6 vs. 23.3 years; \( P < 0.001 \)); 44% of participants (39 of 89) knew little or nothing about DSD, and 37% could not differentiate between transsexualism and intersex; 43% (38 of 89) reported fuller knowledge than provided in the video.

**Participant Decisions and Tendencies**

Thirty-eight participants (43%) opted for sex-assigning surgery for their child; the remainder opted to defer surgery. The relationship between decision and informational video was highly significant: 27 of the 41 participants (66%) who watched the endocrinologist’s medicalized video opted for surgery as against 11 of the 48 participants (23%) who watched the psychologist’s demedicalized video (\( P < 0.001 \)) (Figure 2). Participant conviction in their decision was also significantly associated with the type of information given: those informed by the endocrinologist were more secure and convinced in deciding for than against surgery (5.5 ± 2.5 vs. 7.3 ± 1.8, \( P < 0.028 \)), whereas those informed by the psychologist were more secure in deciding against than for surgery (7.0 ± 2.1 vs. 4.3 ± 2, \( P < 0.001 \)). Neither participants’ gender (\( P = 0.9 \)) nor their prior knowledge (\( P = 0.067 \)) significantly influenced their decision. However, participants rating their knowledge as “detailed” tended to decide against surgery (detailed knowledge and no surgery: 26 of 38 [68%] vs. 25 of 51 [49%]).

**Self-Assessment of Influencing Factors**

All participants believed their decision was based mainly on their own values, opinion, and attitude. On a scale from 1 (no influence) to 4 (strong influence), participants perceived their personal attitude as the main influence (3.6 ± 0.7). Video content (2.8 ± 1) or informant identity (2.3 ± 1), on the other hand, was perceived as having little or no influence (Table 3).

Proponents of surgery judged autonomy to be less important than its opponents (\( P < 0.001 \)). Normality, on the other hand, mattered more for those favoring surgery (\( P < 0.001 \)). The same pattern was found with respect to the video information: significantly more participants judged autonomy as important in their decision after listening to the psychologist than after listening to the endocrinologist (\( P < 0.006 \)), and vice versa for normality (\( P < 0.001 \)).

**Discussion**

Our results delineate the different information behaviors of different professionals and show a strong relationship between informing professional and parental attitude to surgical intervention. An important finding is that subjects (i.e., medical students) were largely unaware of such influences. Contrary to majority participant
perception it was not just their attitude that made
them decide for or against surgery but primarily a
6-minute slot of information. Moreover, our study
found a strong relationship between the desire for
normality plus unambiguous genitalia and expo-
sure to the endocrinologist video as opposed to the
psychologist video.

Our data are in line with the 2005 consensus
statement on DSD recommending further studies
“to evaluate the effectiveness of information
management with regard to timing and content”
[1]. At the same time our results suggest that
current information management concerning
DSD is prone to produce biased decisions.
Although our experimental setting was not meant
to isolate and identify particular factors, our
results help to raise awareness of communication
biases in a particular and hotly debated problem.
Current DSD recommendations and guidelines
show a paradigm shift from a medicalized to a
more demedicalized approach that includes the
kind of nonmedical members, such as psycholo-
gists, social workers, and support groups, who
represent the potentially diverging values and
arguments collected in the initial focus group
(Table 1).

Our data complement current discussions on
the implementation of different approaches to
professionalism in DSD, especially considering
that we collected our data after the consensus
statement and its concomitant call for multidisci-
plinary teams [16]. We identified several problems
and biases, all related to the significant lack of
objective medical and/or quality of life data on

**Table 3** Self-assessed impact of potential determinants of decision for or against surgery on a four-point Likert scale
(1 = no influence, 2 = rather no influence, 3 = rather influential, 4 = influential)

<table>
<thead>
<tr>
<th>Medicalized group (n = 41)</th>
<th>Demedicalized group (n = 48)</th>
</tr>
</thead>
<tbody>
<tr>
<td>For surgery (n = 27)</td>
<td>Against surgery (n = 14)</td>
</tr>
<tr>
<td>Strong influence (mean 3.5–4)</td>
<td>Desire for normality (3.8 ± 0.4)</td>
</tr>
<tr>
<td>Personal attitude (3.7 ± 0.6)</td>
<td>Personal attitude (3.7 ± 0.5)</td>
</tr>
<tr>
<td>Child’s future (3.6 ± 0.5)</td>
<td>Child is healthy (3.6 ± 0.7)</td>
</tr>
<tr>
<td>Desires for normality (3.5 ± 0.7)</td>
<td>Child’s future (3.9 ± 0.9)</td>
</tr>
<tr>
<td>Some influence (mean 2.5–3.4)</td>
<td>Child’s future (3.4 ± 0.9)</td>
</tr>
<tr>
<td>Unambiguous sex (3.1 ± 1)</td>
<td>Social pressure (2.9 ± 1)</td>
</tr>
<tr>
<td>Social pressure (2.7 ± 0.9)</td>
<td>Video information (2.7 ± 1)</td>
</tr>
<tr>
<td>Video information (2.7 ± 0.9)</td>
<td>Time pressure (2.9 ± 1)</td>
</tr>
<tr>
<td>Time pressure (2.6 ± 1)</td>
<td>Prior knowledge (2.8 ± 1)</td>
</tr>
<tr>
<td>Unspecified fears (2.2 ± 1)</td>
<td>Video information (2.6 ± 1.2)</td>
</tr>
<tr>
<td>Rather no influence (mean 1.5–2.4)</td>
<td>Prior knowledge (2.6 ± 1.3)</td>
</tr>
<tr>
<td>Open questions (2.4 ± 0.9)</td>
<td>Risk of surgery (2.8 ± 1)</td>
</tr>
<tr>
<td>Prior knowledge (2.4 ± 1)</td>
<td>Social pressure (2.8 ± 1.1)</td>
</tr>
<tr>
<td>Informing person (2.3 ± 1)</td>
<td>Child’s autonomy (2.1 ± 1)</td>
</tr>
<tr>
<td>Child’s autonomy (2.3 ± 0.9)</td>
<td>Time pressure (2.1 ± 1)</td>
</tr>
<tr>
<td>Child’s health (2.2 ± 1.1)</td>
<td>Informing person (1.9 ± 1)</td>
</tr>
<tr>
<td>Risks of surgery (2 ± 1.1)</td>
<td>Child has a disorder (1.9 ± 0.9)</td>
</tr>
<tr>
<td>Child has a disorder (1.7 ± 1)</td>
<td>Social pressure (1.8 ± 1)</td>
</tr>
<tr>
<td>Weak influence (mean 1–1.4)</td>
<td>Child has a disorder (2.1 ± 0.9)</td>
</tr>
<tr>
<td>Religious beliefs (1.4 ± 0.9)</td>
<td>Religious beliefs (1.2 ± 0.4)</td>
</tr>
</tbody>
</table>

J Sex Med **,***,**—**
long-term outcome with or without genitoplasty in any DSD condition. Although there are studies suggesting better results for surgery in early childhood compared with late childhood or reporting patients and parents who would undergo or choose early surgery in retrospect again [21–23], these data are inappropriate for endorsing genital surgery in childhood in general [5,6]. There is still a great divide between offering parents immediate relief (“I can fix that”) and the residual or subsequent fear, uncertainty, and sense of guilt that parents may experience after surgery, not to mention the potential late sequelae of surgery in the patients themselves [8,11,21,24]. However, criticism of the surgical approach needs to beware of being oversimplistic, given the variations of DSD and the absence of empirical data on the long-term outcome of withholding surgery in early childhood [25].

The absence of empirical data is a serious obstacle when informing parents and patients. But primarily it leads less to an error of judgment than to its impossibility [26]. Furthermore, it is associated with “common sense” processes in which complex questions (target attributes) become replaced by conceptually and associatively related knowledge (heuristic attributes), resulting in an automatic and mostly unconscious search for premises on which to base recommendations and decisions [7,26–28]. The lack of detailed objective criteria, such as a quantifiable probability that a particular patient will lead a happy life after genital surgery, generates systemic weighting biases, giving certain cues (such as standing to urinate or having heterosexual intercourse) too much or too little weight.

Another important source of pressure on the decision-making process lies in framing bias, related to the different frames within which identical information may be presented. Associated with framing bias and of particular ethical importance is parentalism [29], defined as the championing of intervention in support of the parents’ right to decide as a surrogate for their child, even when that decision may largely be induced, precipitately and unconsciously, by a health-care professional, rather than emerging from a balanced, comprehensive, and thought-out process [1,2,8,11,22]. Parentalism may be medicalized or demedicalized. The ethical problem in allowing and fostering parentalism is that it might not only invalidate common expectations of informed consent but also lead to irreversible and inadequately grounded decisions. This takes on great significance if parents and professionals later need to inform the child or adolescent why a certain approach had been chosen [2,11,16]. Our initial focus group discussion identified contrasting attitudes even within a multidisciplinary team, whereas the results of the video experiment showed that decision making requires protection from being based on mistaken, partisan or precipitate premises or swayed by the influence of a perceived authority of a particular professional. Neither the presentation of information in a clear and professional manner nor the current recommendations and guidelines might be sufficient for achieving a holistic, balanced, and collaborative approach [16,17,30–32].

Limitations

The appraisal of our data is subject to severe limitations. Due to the psychological burden of our videos for actual parents we decided to expose only medical students to the experimental setting, as role-playing is already a technique with which they are familiar in their training. Therefore the sample is not necessarily representative of parents in general. Forty-three percent of the participants reported knowing more about DSD than was presented in the video, thus most probably exceeding knowledge among the general public. However, participants’ knowledge had no significant influence on their decision (P = 0.67).

A further source of potential bias is that the two videos did not represent generalizable ways of informing parents. This cannot be done in 6 minutes, nor in a setting where only the professional speaks. The division into medicalized and demedicalized information is neither clear-cut nor generalizable when applied to the complex, multifaceted, and changing environment of multidisciplinary team approaches in supporting children and families with DSD. Moreover, it is simplistic to divide information management into a medicalized (endocrinologist’s) presentation focused on disorders and genetic mutations and the demedicalized (psychologist’s) offer of psychosocial support. Our experimental situation reflected neither the recommendations of the 2005 consensus statement on DSD nor the current advocacy and implementation of specialized multidisciplinary teams [1,16]. Nevertheless our videos were derived from statements made within a “post-consensus” multidisciplinary group pursuing a full consent policy [1]. Our results thereby also point to the possibility of unrecognized or underestimated influences and impacts from earlier times.
In addition, we administered a self-designed questionnaire that had not undergone prior validation. We should also bear in mind that although professional influence was highly significant, there were many participants who did not change their mind in accordance with the medicalized or demedicalized information given to them: 14 of 41 participants (34%) refused surgery after being exposed to medicalized information, whereas 11 of 48 (23%) opted for surgery after being exposed to demedicalized information. Parents, therefore, should not be perceived as passive, innocent bystander. As a consequence a clear demarcation between attitudes, prior knowledge, and mode of counseling is not possible. Further data are needed, which delineate different characteristics of parents and their importance to decision making. Nevertheless, within these limitations, our findings quantify significant biases and identify the central requirements for information management in DSD.

Conclusions

While acknowledging our study’s limitations, we believe that our data confirm the need to supplement current recommendations in four respects:

First, given the absence of hard data, all interventions, whether medical, surgical, or psychosocial, should be part of a comprehensive follow-up (side effects or detrimental outcomes are not exclusive to surgical intervention).

Second, professionals must learn how to establish shared or child- and family-centered decision making. It must include the sharing of complete, honest, and minimally biased information with patients and their families. It must also recognize and build on the strengths of individual children and families as well as empowering them to discover their own strengths [30]. In addition to knowledge of pathogenesis, medical professionals involved in care of patients with DSD should also be educated and trained in psychosocial issues.

Third, all those involved in decision making should be alert to potential heuristic and framing bias. Recent publications point to the importance of parents able to educate their child with DSD about preceding and upcoming decisions for or against certain treatments [33]. Parental competence in the education of their children, however, is related to their own understanding of decisions, which might be severely impaired by biased information.

Fourth, our results point to the importance of additional parameters in making an early distinction between compelling medical problems, such as infection due to an obstructed urinary tract or the increased risk of malignancy associated with dysgenetic gonads, which are properly labeled as disorders, and concomitant variations or differences for which initial demedicalized management, including demedicalized postnatal counseling, may prove superior, even allowing for potential future impairments and the need for continuous reevaluation. As there are no clear-cut lines between disciplines, medicalized and demedicalized approaches can probably go hand in hand within a multidisciplinary team. Our data, however, suggest that multidisciplinary teams should actively reflect their specific mix of demedicalized and medicalized approaches and judiciously select shifts of responsibilities and associated skills (such as when determining whether an endocrinologist, surgeon, or psychosocial support expert should talk to the parents first).

Corresponding Author: Jürg C. Streuli, MD, University of Zürich, Institute of Biomedical Ethics, Pestalozzistrasse 24, 8032 Zürich, Switzerland. Tel: +41 44,634 83 74; Fax: +41 44,634 83 89; E-mail: streuli@ethik.uzh.ch

Conflict of Interest: The authors report no potential conflicts of interest.

Statement of Authorship

Category 1

(a) Conception and Design
Jürg C. Streuli; Yvonne Cavicchia-Balmer

(b) Acquisition of Data
Yvonne Cavicchia-Balmer; Jürg C. Streuli

(c) Analysis and Interpretation of Data
Jürg C. Streuli; Yvonne Cavicchia-Balmer; Effy Vayena; Johannes Huber

Category 2

(a) Drafting the Article
Jürg C. Streuli

(b) Revising It for Intellectual Content
Jürg C. Streuli; Yvonne Cavicchia-Balmer; Effy Vayena; Johannes Huber

Category 3

(a) Final Approval of the Completed Article
Jürg C. Streuli; Yvonne Cavicchia-Balmer; Effy Vayena; Johannes Huber
References


