DSD-Related Attitudes and Beliefs Held by Pediatric Endocrinologists and Urologists: Insights from the North American Disorders/Differences of Sex Development (DSD) Clinician Survey

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Short Title: N Amer DSD Clin Survey: 46,XY Care

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Abbreviations. PES (Pediatric Endocrine Society), SPU (Societies for Pediatric Urology), clinical practice guidelines (CPGs), disorders/differences of sex development (DSD)

Key words:

Knowledge, attitudes, beliefs, Disorders/Differences of sex development, Intersex, sex, gender

DSD Clinician Survey: Survey and Participant Profile Knowledge, Attitudes, and Beliefs

Summary

A survey of pediatric endocrinologists and urologists was administered at three timepoints. Participants were recruited from membership rosters of two North American-based professional societies: the (Lawson-Wilkins) Pediatric Endocrine Society and the Societies for Pediatric Urology. Members were sent invitations to complete the online survey at three timepoints: three years prior to publication of the 2006 "Consensus statement on management of intersex disorders," four years following it, and four years following publication of the 2020 Consensus statement update.

Survey Development

Initial Survey Development

Provisional survey items were generated based on a literature review and focus groups conducted by conference call. Focus groups were convened to identify themes pertinent to the investigation and canvass opinion regarding optimal survey administration format. Focus group participants included 16 junior and senior members of the (Lawson-Wilkins)¹ Pediatric Endocrine Society (PES) and the Societies for Pediatric Urology (SPU) nominated for participation by colleagues who thought their opinions would be particularly informative; a geographically diverse sample was sought. Web-based administration to facilitate recruitment was the consensus of focus group participants. A preliminary survey was pilottested with a subgroup of focus group members with other members checking for comprehensiveness of content coverage and survey response options.

The final version of the Time 1 (T1) Intersex Survey was administered in 2003-2004 and comprised five sections: 1) Case Presentations, 2) Factors Affecting Life Satisfaction, 3) Surgical Informed Consent, 4) Mental Health Services and the DSD Team, and 5) Demographics.

This document provides a detailed description of those completed the second section of the survey (Factors Affecting Life Satisfaction) at 2003-2004, 2010-2011 and 2020 timepoints.

Subsequent Survey Iterations

Items in the T1 survey were edited over time reflecting terminology changes. Use of earlier data collection and analyses guided later data collection. Limited changes were made to item presentation, wording, and order. When considering changes, goals were to limit the extent of changes made to allow for direct comparisons across time.

Revised surveys, T2 and T3, were administered in 2010-2011 and 2020, respectively.

When data collection spanned >1 year, the first year in which data collection occurred is used in the label. These are hereafter referred to as the T1 2003, T2 2010, and T3 2020 surveys.

¹ Founded in 1972, the Lawson Wilkins Pediatric Endocrine Society formally changed its name to the Pediatric Endocrine Society (PES) in 2010.

Edits to items over time:

- Terminology changed from Intersex to DSD. In 2003, intersex applied to the medical condition; it was replaced by disorders of sex development following the 2006 Consensus Statement; by 2020, use of the term intersex re-emerged, but carried a different connotation for some applying more to an identity than a medical condition, per se. Additionally, by 2020 the word disorder was viewed negatively by some who supported the term difference of sex development. Changes in wording were intended to maintain a focus on the same set of medical conditions, despite changes in vernacular used to describe these conditions.
 - T1 2003: "Intersex" and "Intersexuality"
 - T2 2010: "Disorder of Sex Development (DSD)"
 - o T3 2020: "Disorders/differences of sex development (DSD)"
- Use of earlier data collection and analyses to guide later data collection item order
 - o T1 2003 began with case presentations and ended with demographics
 - o T2 2010 began with demographics
 - T3 2020 began with introduction, then demographics; the introduction included a selfadministered eligibility screen and opt-out with pre-populated reasons for opting out
 - T3 2020 included an "other" option for recommended gender of rearing (Case Presentation) and for participant gender (Demographics)

Survey Components

Table 1. Knowledge, attitude, and belief-focused survey components

Section	Contents: Major Components
Introduction	Overview of survey
	Eligibility screener – 2020 only
Demographics	Clinical practice and demographic characteristics
Factors Affecting	Ranking the importance of different factors exerting an influence on adult life
Life Satisfaction	satisfaction among individuals with DSD [Only in 2003]
	Indicating the level of agreement with different statements about human psychosexual development [2003, 2010, and 2020] Predicting experiences of individuals in North American society with regard to mental health, sexual and romantic relations, and marriage [Full sample in 2003; SPU only in 2010]

Table 2. Knowledge, attitude, and belief-focused survey component order

Section	T1: 2003	T2: 2010	T3: 2020
1	Introduction	Introduction	Introduction
2	Factors Affecting Life Satisfaction	Demographics	Demographics
3	Demographics	Factors Affecting Life Satisfaction	Factors Affecting Life Satisfaction

Figure 1. Branching and skip logic used in survey administration



Survey Items

Notes: This survey was administered online; branching and skip logic were employed. Instances of branching and skip logic are shown below with logic described in bracketed sections. Additional changes specific to one or more years are noted in bracketed sections. The order of items presented below reflects that used in the 2020 survey.

Introduction: Eligibility [2020 only]

You were selected to complete this survey due to membership in the [relevant society name is displayed].

Disorders/differences of sex development (DSD) are "congenital conditions in which development of chromosomal, gonadal, or anatomical sex is atypical."

We are aware that not every society member is a practicing clinician working the area of DSD. You can indicate below whether or not you provide care to these patients or if you do not want to participate in the study. By either completing the survey or declining participation, you will not receive any more follow-up reminders to participate.

[2020 only]

Eligibility to participate:

Have you been involved in the clinical management of at least one patient with DSD?

☐ Yes

□ No

- Yes [proceed to Enrollment question]
- [if selected "no"]
 - o Please complete this last question so we can describe non-participants in any publication.
 - Are you (please select all that apply):
 - In clinical practice, with a different patient population (e.g., diabetes...)
 - In research
 - In teaching
 - In admin
 - Retired
 - Other
 - Other text
 - Thank you for your time

[all years]

Enrollment question:

- ☐ Proceed to Survey
- ☐ Decline Participation
- Yes [proceed to survey]
- [2010, 2020: if selected "decline participation"]
 - o So that we can describe non-participants, are you (please select all that apply):
 - In clinical practice with DSD patients
 - In clinical practice, with a different patient population (e.g., diabetes...)
 - In research
 - In teaching
 - In admin
 - Retired
 - Other: [Write-in test response]
 - o Thank you for your time

	GR		

Please describe some characteristics of your practice. The data will be used ONLY for this research study. All responses are strictly confidential. Only averaged data from respondents will be utilized. No information from individual respondents will be provided to any person, group, or agency.

	now many children/adolescents/adults do you see annually who were born with DSD? bout how many children/adolescents/adults do you see annually who were born with intersexuality? cases per year
About I	now many individuals born with DSD have you seen over your entire career? cases over entire career
Please i	indicate the average number of hours per week that you spend in patient care hours per week
	Syour area of specialization? Urology Endocrinology Other:
	describe the community in which your main office/practice is located. Large metropolitan: (Total city and suburban population: 1,000,000 and over) Small metropolitan: (Total city and suburban population: 50,000 - 999,999) Nonmetropolitan/Rural: (Total population: 49,999 or less)
For US	Addresses, please indicate the first digit of your office zip code.
	is your practice located? United States Canada Mexico
	Other

[Note: the following practice location data were not directly collected in 2003 and 2010, but derived from other items (zip code) + recruitment material and added to the dataset]
[2020: if practice location is United States] Where is your practice located: State or District (USA)
[2020: if practice location is Canada] Where is your practice located: Providence or Territory (Canada)
[2020: if practice location is Mexico] Where is your practice located: State (Mexico)
[2020: if practice location is Other] Where is your practice located:
Primary Practice Setting: Solo or two-physician practice Group practice HMO Medical school or hospital-based Other patient care employment: Other non-patient care employment: [2010 only]
[if (2003 or 2010) AND if practice setting – medical school or hospital] If you are medical school or hospital-based, please indicate the percentage of time devoted to the following in your medical school or hospital-based practice: % research % patient care % other
What is your gender? Male Female Other, specify:
In what year were you born?

SECTION 3: FACTORS AFFECTING LIFE SATISFACTION

This section concerns your views, perceptions, and opinions about factors that contribute to positive quality of life outcomes for individuals born with Disorders/Differences of Sex Development (DSD).

[2003] Definitions: Assigned Sex = sex to which child with ambiguous genitalia is assigned. Intersexuality = Diverse class of syndromes characterized by some abnormality or anomaly in the process of physical sex differentiation resulting in a genital phenotype that is not clearly classified as either male or female. Gender Identity = identification of self as either girl/woman, boy/man, or other. Gender Role = behaviors that differ in frequency or level between males and females in this culture and time, such as

Note: The following instructions and rank order items appeared only in 2003

toy play or maternal interest.

•	of factors have been proposed as exerting an influence on adult life satisfaction among individuals born with Rank order the importance of the following factors (1 – most important, 5 – least important):
	Gender identity consistent with assigned sex
	Individual characteristics of the child with intersex (e.g., IQ, temperament/personality)
	Features of the particular intersex syndrome (e.g., karyotype, prenatal sex hormone milieu, accompanying congenital somatic and central nervous system defects, fertility potential)
	Family and community context (e.g., acceptance and support of child versus shame or secrecy, strength of family functioning prior to birth of child with intersex, sharing of age-appropriate medical information with the child, religious or cultural background, region of country and/or rural versus urban)
	Features of the treatment of the intersex syndrome (e.g., repeated genital examinations, repeated genital surgery, erogenous sensitivity of genitalia in adulthood, availability of specialized long-term mental health services, interdisciplinary health care team approach, personal relationship of patient with physicians).

[2010]	Definitions:			
Assign	ed Gender = gender to which	child with disorder of sexua	ıl development (DSD) is assig	gned.
Disord	ers of Sex Development (DSI anatomical sex is atypical.)) = Congenital conditions in	which development of chro	mosomal, gonadal, or
Gende	r Identity = identification of s	self as either girl/woman or l	ooy/man.	
Gende	r Role = behaviors that differ toy play or maternal interes	·	en males and females in this	culture and time, such as
[2020]	Definitions:			
Assign	ed Gender = gender in which	the child is reared.		
Disord	ers/Differences of Sex Devel gonadal, or anatomical sex i		conditions in which develop	ment of chromosomal,
Gende	r Identity = identification of s	self as either girl/woman, bo	y/man, or other.	
Gende	r Role = behaviors that differ toy play or maternal interes		en males and females in this	culture and time, such as
epresei	ndicate your level of agreements your opinion. esponse options are the same	-		·
	☐ Strongly Disagree	□ Disagree	☐ Agree	☐ Strongly Agree
	r identity which is at odds w yish behavior in a genetic m nade.			
	ing likely adult sexual orient	tation of the child with a DS	D is one factor in the gende	r assignment
	ectomy of genetic males wh erone (between 2-6 months		ls to occur prior to the post	natal surge in
	I reconstructive surgery for I ence of at least 3 to 4 cases a		cclusively in "Centers of Exc	ellence" (i.e., centers with

Genital reconstructive surgery for DSD is currently being performed predominantly in "Centers of Excellence."

The adequacy of the genitalia specifically for peno-vaginal intercourse is an important determinant of adult quality of life.

The religious, ethnic, and educational background of the family are important determinants of how well the individual born with DSD will adapt emotionally and socially as an adult.

A child with ambiguous genitalia born to a family that is functioning well is likely to achieve a positive quality of life regardless of the decision regarding gender assignment.

Although surgeons should inform parents of all technically possible options, they should not recommend whether genital surgery should or should not be performed.

To achieve optimal adult quality of life, genetic males born with aphallia should be reared as girls/women.

In the absence of medical necessity (e.g., urinary tract infections, painful erections), hypospadias repair in a genetic male who will be raised as a boy should be postponed until the individual is old enough to provide his own informed assent/consent.

The gender identity and gender-role behavior of genetic males (46, XY) with complete androgen insensitivity (i.e., phenotypically typical female) raised as a girl is identical to that of typical females (46,XX).

The gender identity of a physically healthy genetic male (46,XY) with aphallia who is raised as a girl, and who has received female genitoplasty in the first year of life, is identical to that of typical females (46,XX).

I would recommend that parents of a newborn with DSD contact organizations representing affected individuals (i.e., support and advocacy groups) prior to making any decisions regarding surgery.

[2003, 2010: I would recommend that parents of a newborn with DSD contact organizations representing affected individuals (e.g., Androgen Insensitivity Syndrome Support Group, Hypospadias & Epispadias Association, MAGIC Foundation, etc.) prior to making any decisions regarding surgery.]

Prenatal exposure to androgens is the major determinant of gender identity.

To achieve optimal adult quality of life, genetic males born with cloacal exstrophy should be raised as girls.

Setting aside legal considerations that give broad rights to parents, parents' wishes regarding gender assignment and genital surgery should supersede professional judgement.

Notes: The following items ask how difficult it is to have each of 4 conditions.

In 2003 and 2010, these there are 6 questions per condition; in 2020 the 6 have been collapsed into one item, shown within condition at the end of the 2003 and 2010 items.

In 2010, these questions were not asked of PES members; PES leadership was concerned about the length of the survey.

[2003 & 2010 instructions]

In the following questions, we would like you to predict the experience of the individual under different circumstances. Please check the response option that best represents your opinion.

	esponse options are t	he same for the follow	ing items. For ease of use	e, response options a	re shown once (here at
the top)	□ Never	☐ Rarely	☐ Sometimes	☐ Often	☐ Always
How dif experie		merican society for ma	les with uncorrected sev	ere hypospadias and	d marked chordee to
Positiv	e mental health as a	child			
Positiv	e mental health as a	teenager			
Positiv	e mental health as a	n adult			
Positiv	e genital sexual relat	ions			
Positiv	e romantic relations				
Positiv	e marriage				
	fficult is it in North A	american society for bo ental health and intim	oys/men with uncorrecte ate relationships?	ed severe hypospadia	as and marked
			with a small penis (<-2.5	SD for age adjusted	norms) to experience:
Positiv	e mental health as a	teenager			
Positiv	e mental health as a	n adult			

Positive genital sexual relations
Positive romantic relations
Positive marriage
[2020 only] How difficult is it in North American society for boys/men with a small penis (< -2.5 SD for age adjusted norms) to have positive mental health and intimate relationships?
[2003 & 2010] How difficult is it in North American society for females with an uncorrected very large clitoris (3.0 cm at birth) to experience:
Positive mental health as a child
Positive mental health as a teenager
Positive mental health as an adult
Positive genital sexual relations
Positive romantic relations
Positive marriage
[2020 only] How difficult is it in North American society for girls/women with an uncorrected very large clitoris (3.0 cm at birth) to have positive mental health and intimate relationships?
[2003 & 2010] How difficult is it in North American society for females with a vagina too short to permit full penetration by an average length penis to experience:
Positive mental health as a child
Positive mental health as a teenager

Positive mental health as an adult
Positive genital sexual relations
Positive romantic relations
Positive marriage

[2020 only]

How difficult is it in North American society for girls/women with a vagina too short to permit full penetration by an average length penis to have positive mental health and intimate relationships?

Participants

Recruitment - Procedures

At each timepoint, the research team sought approval from leadership of both the (Lawson Wilkins) Pediatric Endocrine Society (PES) the Societies for Pediatric Urology (SPU) to survey their membership and to provide member rosters that include contact information. Leadership of each society approved the research provided rosters, apart from PES at T3 2020 - citing concerns about burden. As such, at T3 for PES, only those who had previously participated at T1 or T2 were invited for participation. A publicly available directory PES was reviewed to remove names of those who were no longer listed as PES members; no new members were added.

To account for slightly differing timelines for study approval by PES and SPU and to manage the number of individuals targeted for participation and anticipated follow-up reminders, survey invitations were sent in waves, rather than to all participants at once. Invitation letters that included an explanation of the study and survey login instructions were sent society members in 2003-04 (T1), 2010-11 (T2), and 2020 (T3). Participants were also offered a paper-and-pencil version upon request. To optimize recruitment, eligible respondents received up to three follow-up requests to participate. After rates of survey completion dropped to minimal levels for several weeks, final requests for participation to non-responders took the form of a phone call and/or a single-page faxed letter encouraging either participation or to otherwise provide a reason for declining to participate at T1. Through this process, it was learned non-responders were frequently either retired, not in clinical practice, were exclusively involved in research, or not providing care to patients with a DSD. At T2 and T3, final follow-ups took place via email. Additionally, a screening and opt-out survey was added to the beginning of the T3 survey to simplify and streamline efforts at identifying participant (in)eligibility and reduce burden associated with follow-ups contacts for non-responders.

EligibilityCommon across all timepoints: Current members of either PES or SPU

Table 3. Eligibility requirements unique to specific timepoint:

		T1	T2	T3
Clinical	Is currently providing care for patients with intersex/DSD conditions	Х	Х	Х
Management	Has not provided care for patients with intersex/DSD conditions, but	Χ		
	are in a position where they could, potentially, do so now or in the			
	future or otherwise influence clinical care			
	Provided care for patients with intersex/DSD conditions in past only ¹	Х	Х	Χ
Professional	Pediatric Endocrinologist, Other Endocrinologist	Х	Χ	Χ
Background	Pediatric Urologist, Other Urologist	Х	Χ	Χ
	Other professional degree (eg, PhD)	Х		
Practice	United States	Х	Χ	Χ
Location	Canada	Х	Χ	Χ
	Mexico	Χ	Χ	Χ
	Other		Х	

¹ Emeriti were, initially, included in the T1 survey; a combination of difficulty reaching these potential participants due to missing or inaccurate contact information, low participation rates, and feedback from several who targeted for participation that they are retired / unable to provide valid input (in their estimation) lead to discontinuing recruitment of emeriti as the project progressed.

Participants were promised confidentially of their responses; procedures were approved by the authors' Institutional Review Boards at the University at Buffalo School of Medicine and Biomedical Sciences (T1 2003) and University of Michigan Medical School (T2 2010 and T3 2020).

Participation Rates and Demographics

Given small differences in eligibility criteria for recruitment, participation rates are calculated using the most restrictive eligibility criteria.

Table 4. Participant ascertainment, recruitment, and participation

Table 4. Farticipant ascertainment	T1 2003 T2 2010				T3 2020		
	PES	SPU	PES	SPU	PES	SPU	
Names in Directory	764	263	868	237	494 ⁶	354	
Ineligible ¹							
Co-I, FG, or PT participant ²	14	9	12	8	10	8	
Emeriti	117						
Retired	11	5	10	6	10	21	
Deceased	2		1				
No DSD patients	34	6	29	3	15	37	
No clinical practice	16	1	16		4	3	
Practice outside N. America	1		13		8	2	
Other; >1 criteria met	53	52	10	2	13	2	
Eligible, invited, sample	516	190	777	218	434	281	
Participated	289	123	319	114	111	144	
Participation rate	56.0%	64.7%	41.1%	52.3%	25.6%	51.2%	
Logged in / consented only ³					5	10	
Declined participation ⁴	48	10	27	3	5	5	
No Response	179	57	429	100	311	117	
Eligible but not invited	0	0	1	1	0	0	
No contact information	0	0	1	0	2	5	

¹ Ineligibility was determined at multiple stages. Determinations were made prior to sending survey invitations to members; for others, it occurred after invitations were sent. It is possible that some of those for whom no responses are recorded are ineligible.

²Co-investigators, focus group members, and pilot test participants involved in the design of this project.

³ At T3, several targeted participants had logged into the survey and completed portions of the screening survey or demographics, but did not provide responses to items in any other section; this was not possible in earlier years.

⁴ A common reason cited for declining participation was being "too busy."

⁵ Reflects an error in recruitment

⁶ Only current members of PES who had participated in the past were included in the 2020 PES sample; this does not represent the total number of names listed in the PES directory.

Survey Completeness

In line with research ethics, participants were free to skip over questions they did not wish to answer and discontinue participation at any time. As such, not all surveys have 100% of items answered – the use of branching and skip logic notwithstanding.

Given the order in which major sections of the survey were presented and order of items within sections (see Survey Components, above), discontinuation disproportionately affected response rates of individual items depending on the year of administration.

Table 5. Participant demographics

B. C. Proposition	T1 2003				T2 2010				T3 2020			
	PES		SPU		PES		SPU		PES		SPU	
	n	%	n	%	n	%	n	%	n	%	n	%
Sex												
• Male	174	60.20%	117	95.10%	159	49.80%	108	94.70%	57	51.40%	123	85.40%
• Female	115	39.80%	6	4.90%	160	50.20%	6	5.30%	54	48.60%	21	14.60%
• Other¹			-						1	0.8	0	0
Practice Community ²												
 Large Metropolitan 	173	60.70%	86	71.70%	199	62.40%	79	69.30%	76	68.50%	108	75.00%
Small Metropolitan	107	37.50%	33	27.50%	120	37.60%	33	28.90%	34	30.60%	34	23.60%
Nonmetropolitan / Rural	5	1.80%	1	0.80%			2	1.80%	1	0.90%	2	1.40%
Practice Country												
 United States 	271	93.80%	116	94.30%	301	94.40%	109	95.60%	103	92.80%	138	95.80%
Canada	18	6.20%	7	5.70%	18	5.60%	5	4.40%	8	7.20%	6	4.20%
Practice Setting ³												
Medical School or Hospital	212	76.8	78	65.5	247	77.40%	71	62.30%	93	83.80%	105	72.90%
Solo or 2-physician Practice	22	8.0	16	13.4	72	22.60%	43	37.70%	5	4.50%	6	4.20%
Group Practice	33	12.0	24	20.2					11	9.90%	30	20.80%
• HMO	9	3.3	1	0.8					1	0.90%	3	2.10%
Other	0	0	0	0					1	0.90%	0	0.00%
	mean	SD	mean	SD	mean	SD	mean	SD	mean	SD	mean	SD
Year of Birth	1950.9	9.1	1951.5	8.9	1960.0	10.8	1955.7	8.1	1960.7	9.2	1965.9	9.4
Cases Seen												
In Past Year	10.1	25.4	8.3	8.6	14.7	57.3	11.0	16.2	13.8	21.7	11.1	18.0
Over Career	65.9	87.8	91.3	121.8	84.8	135.0	131.9	230.8	99.5	155.2	133.8	426.6
Hours/Week in Patient Care	28.0	15.6	52.6	15.5	28.4	15.6	50.0	15.3	26.2	12.9	46.1	13.3
Proportion of Time Spent in ⁴												
Research	25.0	23.4	11.5	11.0	27.7	27.4	10.7	9.9				
Patient Care	55.8	25.5	77.6	13.8	56.3	27.2	77.8	15.5				
Other	19.2	16.6	10.9	11.2	16.3	14.8	11.4	12.6				

Note: Not all participants answered every item; percentages are calculated based on number of valid responses to each item rather than on cohort size ¹ "Other" was included as a response option at T3 only; ² Small Metropolitan and Nonmetropolitan / Rural were collapsed for PES members at T2; ³ Practice Setting was parsed into 2 categories at T2; ⁴ Items was asked only of those who indicated working in a "Medical School of Hospital" Practice setting; Item was not included at T3