



Experiences and preferences of women with CAH and parents about disclosure of childhood surgery

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Keywords

Adrenal hyperplasia; Congenital; Differences of sex development; Patient reported outcome measures

Abbreviations

Congenital adrenal hyperplasia, CAH; Female genital restorative surgery, FGRS; Differences of sexual development, DSD; Human immunodeficiency virus, HIV

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Summary

Introduction

Complete disclosure of childhood genital surgery to patients with congenital adrenal hyperplasia (CAH) is a critical part of CAH care. There are no guidelines or uniform recommendations on the timing and content of surgical disclosure discussions.

Objective

Our objective was to describe the experiences and preferences of females with CAH and parents of females with CAH who underwent childhood genital surgery regarding surgical disclosure.

Methods

We conducted an anonymous cross-sectional online survey of females with CAH (46XX, ≥ 16 years [y] old) and parents of females with CAH who underwent genital surgery before age 4y in North America. Participants reported experiences, preferences, and advice about initial ("first time you were told") and complete disclosure ("told all details"). Non-parametric statistics and qualitative analysis were used.

Results

Participants included 59 females with CAH (median age: 37y, 92% White, 93% non-Hispanic) and 41

parents (median: 36y, 85% White, 93% non-Hispanic, daughter median: 26y). The 76% of females who received complete disclosure were younger (median age: 33y) and underwent surgery more recently (median decade: 1980s) than the 14% who received only initial disclosure (median: 47y, 1970s) and the 10% who did not receive any disclosure (median: 60y, 1960s, $p = 0.0003$, **Summary Figure**). Females reported median ages of initial and complete disclosure as 7-10y and 11-13y, respectively. Disclosure was preferred by 98% of females with initial disclosure by age 14y and complete disclosure by 18y. Parents reported similar findings. Most disclosures were by mothers (initial: 82%, complete: 64%). Doctors were more involved in complete vs. initial disclosures (complete: 47%, initial: 13%, $p < 0.001$). Qualitative analysis of advice about surgical disclosure revealed 8 themes.

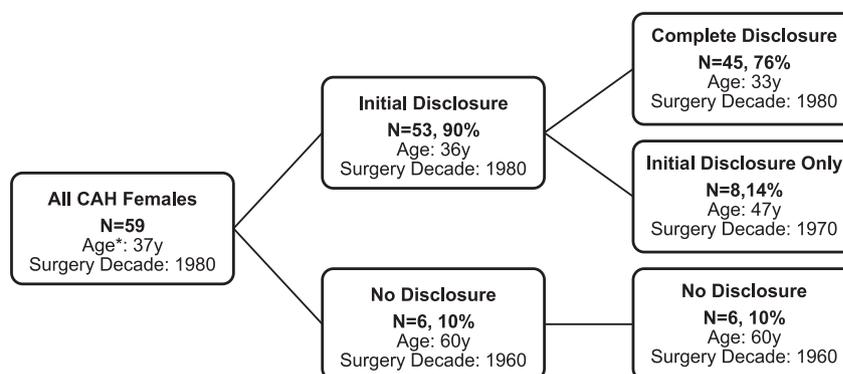
Conclusions

Disclosure of childhood genital surgery to women with CAH has increased over time. Although timing of disclosure varied, women preferred disclosure, and that it be initiated before age 14y and completed by age 18y.

¹ A complete list of study group members appears in the [Appendix](#).

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*Median age at time of survey participation

Summary figure Surgical disclosure of females with CAH

Introduction

The debate regarding genital surgery in childhood for patients with congenital adrenal hyperplasia (CAH) continues. Regardless of changes to legislation or individual institutional policies, historically, many females with CAH (46 XX) have undergone female genital restorative surgery (FGRS) in childhood, typically in the first year of life [1–3]. Although most females with CAH and parents prefer surgery in early childhood [4,5], the decision to proceed with surgery is complex and raises numerous issues [6]. While a comprehensive discussion regarding these issues is beyond the scope of this paper, issues include parents' concerns of subsequently disclosing to their daughters the decision to pursue early genital surgery.

Complete disclosure of genital surgery to people with CAH has become a crucial principle of CAH care. The 2006 Differences of Sexual Development (DSD) Consensus emphasized transparency and open communication between parents, patients, and physicians [7]. This has been reiterated by the Sexuality Information and Education Council of the United States and the CARES foundation, a large patient and family advocacy group for CAH [8,9]. Nearly all parents whose daughters with CAH underwent surgery (94%) agreed on the importance of disclosure [6]. However, there are no guidelines or uniformity of published recommendations on the timing of surgical disclosure, and no publications capture the voices of patients [6,9,10].

We aimed to describe the experiences and preferences of females with CAH and parents of females with CAH who underwent childhood genital surgery regarding surgical disclosure. We sought to determine the ages at which disclosure had occurred and preferred timing according to both females and parents. Secondarily, we sought the recommendations of females and parents on disclosure discussions. We hypothesized that females with CAH would prefer to receive surgical disclosure, and for it to occur before adulthood.

Methods

The Life with CAH Study Group was established in 2019 to research patient-/parent-centered CAH topics. As previously described [11], our methodology included an Institutional Review Board-approved, anonymous online survey. We included CAH females (46XX) ≥ 16 years old (y) who had undergone FGRS before 4y and parents of CAH females who underwent FGRS in the United States and Canada. Any female and parent dyads were not paired. Respondents were included if they answered the statement, "I was born ..." by selecting "Female (46XX)." We did not gather data on gender; thus our usage of "female" or "women" refers to the sex of the respondents.

Questionnaire development and distribution

This research topic was prioritized by people directly affected by CAH, rather than clinicians or researchers. The questionnaire was drafted by an interdisciplinary international medical team (endocrinologists, psychologist, ethicist, sexual/reproductive health researcher, urologists and nurses involved in CAH care, see **Acknowledgements**). Phrasing was adjusted based on feedback from females with CAH and parents to maximize comprehension and face validity. As described previously [11], multiple precautions were used to safeguard the integrity of the questionnaire during questionnaire design, administration, and analysis [12].

Between 2019 and 2020, the questionnaire (35% response rate) was serially distributed via email 1–2 months apart to 5 groups: 4 multidisciplinary CAH clinics (3 in the United States and 1 in Canada, see **Acknowledgements**) and CARES Foundation members. It was accessible for 1 week using a time-sensitive password-protected link. A follow-up invitation was emailed a month later with another individualized, password-protected link available for 1 week. During analysis, serial repeating answers and IP

addresses were screened to detect data dumping (none observed).

Questionnaire content

Questionnaires were similar for CAH females and parents. In addition to demographics, participants were asked about surgical history including the age of first genital surgery (see **Supplement**). Questions regarding disclosure were introduced with: "Many women with CAH as well as their parents have shared with us that they have a lot of questions about how and when to discuss any genital surgery that happened in childhood. We would like to learn from your experience to help other women and families." *Initial disclosure* was defined as being "first told you (your child) had surgery on your (your child's) genitalia." *Complete disclosure* was defined as being "told all the details about the surgery on your (your child's) genitalia." Participants were asked the age categories at which initial and complete disclosure occurred, and what ages they would have preferred ("Looking back, at what age would it have been best for you to be told ..."). We avoided using discrete ages in years since these would be likely to imprecisely recollect many years later. Ages were also paired with expected schooling levels to aid in recollection. Age options included: 6y or younger (before elementary school), 7-10y (elementary school), 11-13y (middle school), 14-17y (high school), 18y or older.

Additional questions addressed specific details of disclosure conversations. Participants were asked who initiated the disclosure conversations, who provided disclosure, and who they would have preferred provide disclosure. Participants were asked, "Once you had some time to get used to this information, how did hearing about your surgery affect your relationship with yourself/parent(s)/doctors and medical team" (5-point Likert scale: positive, neutral, negative, mixed [both positive and negative], and not sure/prefer not to answer).

Statistics

Non-parametric statistical tests were used: Fisher's exact for categorical and Wilcoxon rank sum for continuous data. A critical $p = 0.05$ was used to denote significance (software: Stata, StataCorp, College Station, TX, USA).

Qualitative analysis

A research team experienced in qualitative analysis and CAH care ($n=3$) used a thematic approach to analyze the content of open-ended answers, examining emergent categories of advice regarding disclosure conversations. All answers were organized into 3 separate spreadsheets: advice for females with CAH from females with CAH, advice for parents of females with CAH from females with CAH, and advice for parents of females with CAH from parents of females with CAH. Responses with multiple sentences or statements were separated for unique analysis. A total of 349 statements were first categorized by 1 researcher and then examined and discussed with 2 other researchers resulting in 8 emergent themes.

Results

Females with CAH

Participants included 59 females with median age 37y (IQR: 12–29, **Table 1**). The majority were White (92%) and non-Hispanic (93%). Over half of women had earned an advanced degree and 63% were employed full-time.

Ninety percent of females with CAH received disclosure. Specifically, 76% received complete disclosure and 14% received initial disclosure only (**Summary Figure**). The remaining 10% of women reported they did not receive formal disclosure and had learned about their surgery through other means. How they learned about their surgery was not formally captured in this study. In the qualitative

Table 1 Participant demographics.

Demographic	Females with CAH n = 59	Parents of females with CAH n = 41
Age		
Mean (range)	37 (16–64)	35 (23–50)
Median (IQR)	37 (27–44)	36 (30–38)
Race of person with CAH		
White	54 (92%)	35 (85%)
Black	1 (2%)	0 (0%)
Asian	1 (2%)	0 (0%)
Multiple	2 (3%)	3 (7%)
Other	0 (0%)	1 (2%)
Prefer not to answer	1 (2%)	1 (2%)
Hispanic	4 (7%)	3 (7%)
Annual household income		
< \$20k	5 (9%)	0 (0%)
\$20–39,999	14 (24%)	2 (5%)
\$40–59,999	8 (14%)	3 (7%)
\$60–79,999	5 (9%)	2 (5%)
\$80–100k	9 (15%)	5 (12%)
\$100k	11 (19%)	21 (51%)
Not sure	1 (2%)	2 (5%)
Prefer not to answer	6 (10%)	6 (15%)
Education		
< High school	2 (3%)	0 (0%)
High school/GED	5 (9%)	5 (12%)
Some college	10 (17%)	6 (15%)
Associates	12 (20%)	0 (0%)
Bachelors	20 (34%)	17 (42%)
Graduate	10 (17%)	13 (32%)
Employment		
Full-time	37 (63%)	21 (51%)
Part-time	4 (7%)	11 (27%)
Unemployed, looking	5 (9%)	2 (5%)
Unemployed, not looking	3 (5%)	5 (12%)
Disability	7 (12%)	2 (5%)
Prefer not to answer	3 (5%)	0 (0%)

feedback, one female reported her endocrinologist mentioning it, and a second female reported being told by an older sibling that she was “born with a penis.”

There was a significant age difference between the three disclosure groups. Those who received complete disclosure were the youngest at the time of survey participation (median age 33y), followed by those who only received initial disclosure (47y) and those who never received disclosure (60y, $p < 0.001$, **Summary Figure**). A similar pattern was observed regarding when genital surgery occurred. Females who received complete disclosure underwent surgery in the 1980s (median), those who received only initial disclosure underwent surgery in the 1970s, and those who never received disclosure underwent surgery in the 1960s. Overall, 98% of patients preferred receiving disclosure of their childhood genital surgery.

Of 53 women who received initial disclosure, the median age of initial disclosure was 7–10y with 38% receiving it in this age range (**Table 2**). This was followed by initial disclosure at 11–13y (26%), 14–17 (17%), and ≤ 6 y (15%). Only 2% received initial disclosure in adulthood. Most women (71%) who received initial disclosure when they were 13y and younger preferred the age at which they received it. Conversely, 70% of those receiving initial disclosure when they were 14y and older preferred initial disclosure at an earlier age ($p = 0.02$).

The median age of complete disclosure was 11–13y (**Table 2**). Females received complete disclosure most frequently at 14–17y (29%), followed by 11–13y (24%), 7–10y (18%), ≤ 6 y (13%), and ≥ 18 y (13%). Most females with CAH (97%) who received complete disclosure before 18y preferred the age at which it was provided, 3% preferred an earlier age, and none preferred complete disclosure after 18y. In contrast, 66% of females who received complete disclosure as adults preferred complete disclosure before 18y ($p = 0.001$).

Mothers typically initiated disclosure conversations (initial: 76%, complete: 53%), with doctors being the second

more common initiator (initial: 24%, complete: 36%). Some females reported initiating the conversations themselves (initial: 11%, complete: 20%). Initial and complete disclosures were most often subsequently accomplished by mothers (initial: 82%, complete 64%). Doctors had an increased role in disclosure conversations from initial (13%) to complete disclosure (47%, $p < 0.001$). This coincided with females’ preferences for disclosure to involve mothers (78%) and physicians (47%). Fathers were rarely involved in disclosure conversations (initial: 2%, complete: 4%), matching the preferences of surveyed females (4%).

Females reported that the overall disclosure experience had predominately neutral or mixed effects on their relationships with their parents (68%) and doctors (62%). Disclosure had positive effects on 18% of parent and 20% physician relationships with the patient. Fewer females believed disclosure had negative effects on their relationships with parents (7%) and doctors (13%).

Parents of females with CAH

The median age of 41 parents of females with CAH was 36y (IQR: 30–38, **Table 1**). Most parents were mothers (85%), followed by fathers (12%) and both parents (2%). At the time of survey participation, the median age of their daughters with CAH was 22y (IQR 18–26).

Parental results mirrored those of females with CAH, with the median age of initial disclosure at 7–10y and complete disclosure at 11–13y (**Table 3**). Thirty-eight (93%) of parents had provided initial disclosure. Two parents had not yet initiated disclosure believing their daughter to be too young at 12 and 17y, and one parent did not provide a reason (daughter 13y). All initial disclosures occurred by 17y, and 90% of these parents preferred the age at which disclosure was provided. Ninety-seven percent of parents completed disclosure, all before adulthood. Of those who completed disclosure in their child’s teenage years (11–17y), 95% preferred the age

Table 2 Self-reported timing and preferences of disclosure of childhood genital surgery among females with CAH.

Self-reported Actual Age (years)	Preferred Age (years)						Never	Unsure
	≤ 6	7–10	11–13	14–17	≥ 18			
Initial disclosure (N = 53)								
≤ 6	8 (15%)	7 (88%)	0 (0%)	1 (12%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
7–10	20 (38%)	1 (5%)	17 (85%)	2 (10%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
11–13	14 (26%)	1 (7%)	2 (14%)	10 (71%)	1 (72%)	0 (0%)	0 (0%)	0 (0%)
14–17	9 (17%)	1 (11%)	1 (11%)	4 (45%)	3 (33%)	0 (0%)	0 (0%)	0 (0%)
≥ 18	1 (2%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (100%)	0 (0%)	0 (0%)
Unsure	1 (2%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (100%)
Total	53 (100%)	10 (19%)	20 (38%)	17 (32%)	4 (7%)	1 (2%)	0 (0%)	1 (2%)
Complete disclosure (N = 45)								
≤ 6	6 (13%)	5 (83%)	0 (0%)	1 (17%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
7–10	8 (18%)	0 (0%)	8 (100%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
11–13	11 (24%)	0 (0%)	0 (0%)	9 (82%)	2 (18%)	0 (0%)	0 (0%)	0 (0%)
14–17	13 (29%)	0 (0%)	0 (0%)	1 (8%)	12 (92%)	0 (0%)	0 (0%)	0 (0%)
≥ 18	6 (13%)	0 (0%)	1 (17%)	3 (50.0%)	0 (0%)	2 (33%)	0 (0%)	0 (0%)
Unsure	1 (2%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (100%)
Total	45 (100%)	5 (11%)	9 (20%)	14 (31%)	14 (31%)	2 (4%)	0 (0%)	1 (2%)

Bolded cells indicate an exact agreement between preferred and actual ages reported.

Table 3 Parent-reported timing and preferences of disclosure of childhood genital surgery among females with CAH.

Self-reported Actual Age (years)	Preferred Age (years)						Never	Unsure
	≤6	7–10	11–13	14–17	≥18			
Initial disclosure (N = 41)								
≤6	19 (46%)	15 (79%)	0 (0%)	1 (5%)	0 (0%)	0 (0%)	0 (0%)	3 (16%)
7–10	13 (32%)	0 (0%)	13 (100%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
11–13	3 (7%)	0 (0%)	0 (0%)	3 (100%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
14–17	3 (7%)	0 (0%)	0 (0%)	0 (0%)	3 (100%)	0 (0%)	0 (0%)	0 (0%)
≥18	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Not yet	2 (5%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (50%)	1 (50%)
Never	1 (2%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (100%)
Total	41 (100%)	15 (37%)	13 (32%)	4 (10%)	3 (7%)	0 (0%)	1 (2%)	5 (12%)
Complete disclosure (N = 38)								
≤6	8 (21%)	5 (63%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	3 (34%)
7–10	9 (24%)	0 (0%)	6 (67%)	3 (33%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
11–13	10 (26%)	1 (10%)	0 (0%)	9 (90%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
14–17	10 (26%)	0 (0%)	0 (0%)	0 (0%)	10 (100%)	0 (0%)	0 (0%)	0 (0%)
≥18	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Not yet	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Never	1 (3%)	0 (0%)	0 (0%)	0 (0%)	1 (100%)	0 (0%)	0 (0%)	0 (0%)
Total	38 (100%)	6 (16%)	6 (16%)	12 (32%)	11 (29%)	0 (0%)	0 (0%)	3 (8%)

Bolded cells indicate an exact agreement between preferred and actual ages reported.

disclosure was completed, versus 64% of parents who completed disclosure at younger ages ($p = 0.005$).

Qualitative results

Eight themes emerged from written feedback for females with CAH and parents on the nature of surgical disclosure (Table 4). (1) Disclosure should be thorough and honest. If parents create an open dialogue, it will encourage their daughters to ask questions. (2) Tailor conversations to your daughter's age and level of development. (3) Normalize your daughter's diagnosis and surgery. Females should be reassured that they can lead normal lives and that the decision for surgery was made to preserve or improve bodily functions. (4) Explain the surgery in terms of anatomy and function. Disclosure should be medically oriented, use anatomic images (participants did not specify preference for photographs versus diagrams) and focus on bodily functions. (5) Set a calm tone and focus on facts. (6) Focus on acceptance and body positivity. By using a positive tone, parents will ensure that their children feel loved and respected. (7) Go at your daughter's pace over time. Disclosure is a process spanning multiple conversations over time. (8) Avoid terms that highlight abnormality or masculinization. Aside from an emphasis on the use of anatomical names (Theme 4), participants did not recommend any other specific terms that should be used.

Discussion

The decision to proceed or not proceed with FGSR for young 46XX girls with CAH is complex [7,13,14]. Parents enter the decision with multiple concerns including when and how to subsequently disclose surgery to their daughters [6]. Ninety percent of parents do not receive any guidance on

disclosure of urologic surgery, increasing parental anxiety around disclosure conversations [10,15]. To help counsel parents, our study provides missing and much needed perspectives of women with CAH on the timing and effects of disclosure of childhood surgery.

Our findings indicate that women with CAH and parents of daughters with CAH prefer that childhood FGSR be completely disclosed to children. Women prefer for surgical disclosure to be initiated by 14y and be completed by 18y. However, as the qualitative data indicates, one should regard disclosure as a series of on-going conversations rather than deadlines [13,16]. Women with CAH cautioned parents not to push the conversation if their daughters do not seem open, but still encouraged parents to keep engaging their daughters in conversation. As children with CAH age and become more sexually mature, their diagnosis will manifest new issues that will further dictate disclosure discussions.

Historically, disclosure of genital surgery in children with CAH or DSD diagnoses was discouraged [13,17]. A paternalistic approach underestimated both parents' and patients' desire to discuss and understand their conditions and surgeries [18]. With today's welcome emphasis on shared decision-making and transparency, full disclosure regarding childhood FGSR is a crucial part of patients' healthcare journeys [6,13,18].

This evolution was reflected in our results. Women who never received disclosure were the oldest group in their 60s at the time of study participation and underwent surgery in the 1960s. Comparatively, women who received complete disclosure were the youngest group: they were in their 30s and received surgery in the 1980s. One would expect this trend to continue, especially given high anticipated disclosure rates reported by parents [6,10], providing reassuring proof of open communication in current CAH care.

Table 4 Advice with representative quotes from females with CAH and parents of females with CAH on discussing genital surgery.

Theme	Advice for girls and women from other females with CAH	Advice for parents from females with CAH	Advice for parents from other parents of females with CAH
Be thorough and honest	<p>"Be open and ask a ton of questions"</p> <p>"Ask many questions, don't be shy"</p> <p>"Ask as many questions as you would like"</p>	<p>"Be completely honest and open"</p> <p>"Be very open and never avoid questions"</p> <p>"Explain why you made the decision"</p>	<p>"Be honest and upfront"</p> <p>"Answer questions honestly and directly"</p> <p>"Tell her everything"</p>
Tailor conversations to your daughter's age and level of development	<p>"Ask questions if something doesn't make sense"</p>	<p>"Tell her when she's ready to understand. There is no age that is perfect for everyone"</p> <p>"Explain it in terms they will understand"</p> <p>"Use age-appropriate language"</p>	<p>"Talk to her in an age-appropriate way early"</p> <p>"Explain in terms she can understand"</p> <p>"Share age-appropriate facts"</p>
Normalize your daughter's diagnosis and surgery	<p>"You can live a normal life like any other young woman"</p> <p>"You are normal but special"</p> <p>"Talk to other women with CAH"</p>	<p>"Explain that they aren't different from other girls"</p> <p>"This was for medical reasons not appearance"</p> <p>"Make sure your daughter knows that the surgery does not impact who she is as a person"</p>	<p>"Tell her children have surgery for many reasons due to birth differences"</p> <p>"Everyone has a different body"</p> <p>"Tell them that they can still have a normal life like everyone else"</p>
Explain the surgery in terms of anatomy and function	<p>"Ask for visual aids to help understand your anatomy and how it functions"</p> <p>"Learn about your own body and its own functions so you are comfortable in a sexual relationship"</p>	<p>"Use correct anatomical words"</p> <p>"Use diagrams to show her what female genitalia looks like and what the surgery improved"</p> <p>"The best way my father/doctor explained to me was that I was a female inside, with ovaries and a female reproductive system, but with an exterior malformation"</p> <p>"Show pictures before and after [surgery] to understand the decision"</p>	<p>"Explain it in terms of wanting her to have the best possible genital sensation and function"</p> <p>"This is what she needed for her body to function without medical issues"</p> <p>"I showed her the sketch the surgeon drew to explain what he was doing"</p> <p>"We are not at all choosing the gender. We are simply correcting the genital malformation"</p>
Set a calm tone and focus on facts	<p>"Try not to over-exaggerate the situation"</p> <p>"Don't feel ashamed or embarrassed"</p> <p>"Try to stay calm"</p>	<p>"I would have appreciated my parents to have been blunt rather than sugar-coating it"</p> <p>"Don't make a big deal out of it"</p> <p>"Letting it be casual made it seem more casual to me and therefore wasn't something I overthought"</p>	<p>"Talk about it matter-of-factly, calm, and positive"</p> <p>"Be positive and factual"</p> <p>"Don't be emotional"</p>
Focus on acceptance and body positivity	<p>"Your parents love you and made the choice they believed was right for you"</p> <p>"Your body is normal and won't be weird to a sexual partner"</p>	<p>"Be open, loving, and understanding"</p> <p>"Explain that most partners do not know the difference"</p> <p>"Make sure your daughter knows that the surgery"</p>	<p>"Treat your child with unconditional love and respect"</p> <p>"Tell her that you love her and want what's best for her"</p>

Table 4 (continued)

Theme	Advice for girls and women from other females with CAH	Advice for parents from females with CAH	Advice for parents from other parents of females with CAH
Go at your daughter's pace over time	<p>"Accept yourself for who you are"</p> <p>"Ask any questions if you need to even if it takes you time to digest the information, and you have to ask later"</p> <p>"Take your time to process"</p> <p>"Listen and keep an open mind"</p>	<p>does not impact who she is as a person"</p> <p>"Let them know you are there for any other questions they have"</p> <p>"Don't push talking about it if your daughter doesn't seem open to discussing"</p> <p>"She won't understand much at the time, but keep explaining it"</p>	<p>"Let them know that it doesn't change anything about them"</p> <p>"Keep communicating as they grow"</p> <p>"Talk about it more than once so she has time to think about questions"</p> <p>"Follow up a few weeks later to make sure there are no questions"</p> <p>"They may not understand all that is explained and the older they become, they may want to revisit the details to better understand"</p>
Avoid these terms		<p>Normalizing surgery</p> <p>Normal</p> <p>Abnormal</p> <p>Not normal</p> <p>Ambiguous</p>	<p>Hermaphrodite</p> <p>Intersex</p> <p>Masculinized</p> <p>Male</p> <p>Boy</p> <p>Deformity</p> <p>Normal</p> <p>Abnormal</p> <p>Penis</p>

Medical disclosure in childhood has many known positive effects. The robust literature on childhood disclosure of human immunodeficiency virus (HIV) emphasizes the positive mental, emotional, social, and physical effects on children [16]. Although children may experience immediate confusion and anger, these dissipate and are replaced by more positive emotions of normalcy and acceptance as quickly as six months after HIV disclosure [19]. Parental concerns that HIV disclosure could negatively affect their relationships with their children have been negated as children did not report any change in relationship quality [20]. Similarly, in our study the majority of female respondents reported positive, neutral, or mixed effects on both their relationships with their parents and their doctors. Those diagnosed with DSD in childhood reported greater acceptance of their diagnoses and their own bodies compared to those who received their diagnoses as adults [21]. Withholding important medical information from children can have negative long-term health effects, lowering adherence with medical care and follow-up with clinicians in adulthood [16,18].

Although most parents understand the importance of disclosure of urologic surgery, parents report variable expected ages for disclosure ranging from toddler to young adulthood [10]. Despite uncertainty, parents, pediatricians, and psychologists agree that the timing for these complex conversations should be individualized to the patient's development and comprehension [10,13,16]. Sexuality comprehension is a process: elementary school-aged children can be expected to accurately identify genitalia,

while teenagers should have a deeper understanding of sex [8]. These milestones match our results with the median ages of initial disclosure at 7-10y and complete disclosure at 11-13y.

As outlined by the National Sex Education Standards [8], discussions with children should be medically accurate and evidence based. This was echoed by the study participants who recommended parents use medical phrasing and diagrams, including anatomical variations and the use of surgery to preserve or improve bodily functions. As suggested by our participants, terminology should be factual, avoiding terms that masculinize their anatomy or highlight abnormalities.

Our study has several limitations. Firstly, most participants were non-Hispanic Whites with higher-education degrees and annual household incomes in the higher brackets. Recruited participants were also actively involved in advocacy groups or followed at tertiary care centers. Thus, these results may not be generalizable to minority or lower socioeconomic status groups, or those not supported by advocacy groups or without regular healthcare providers. Similarly, our results may be affected by self-selection bias. As data was not gathered for nonrespondents, the full effect of this bias cannot be determined.

The recommendations provided by participants represent individualized opinions. Disclosure varies from culture [16] and our findings are limited to North America. Any guidelines should emphasize that disclosure discussions are highly personal and are best individualized to the specific values and preferences of the patients, their

families, and available resources. Future studies would need to recruit a greater spectrum of participants to capture the wider breath of patient experiences.

Secondly, online participants were anonymous. While this may have allowed for unrestricted honesty, we were unable to correlate these findings with patient-specific information such as objective medical or surgical history (e.g., genital anatomy, complications, or need for additional surgeries). We cannot determine whether such factors would alter the timing or effects of disclosure. We could not follow participants longitudinally to understand the effects of disclosure over time. We could not link responses of females to their parents if both participated.

Additionally, without knowledge of the exact number of or actual nature of disclosure conversations, we needed to rely on the respondents' reports. For example, a patient and parent may have differing opinions on the completeness of disclosure. For those reporting never having received disclosure, it was sometimes unclear how they learned of their surgeries. Although our data included effects on interpersonal relationships, we did not evaluate the emotional effects of disclosure or the role that psychosocial support played in the process. Patients, parents, and clinicians could benefit from a better understanding of the psychosocial needs of patients with CAH.

Respondents provided information based on their memories of disclosure, predisposing their responses to potential recall bias. We also found that many respondents preferred the age at which they received disclosure, which could, at least partially, be due to confirmation bias with respondents wanting to validate their own lived experiences. However, we did find that for females who had received disclosure past a certain age (initial: >14y, complete: >17y), they preferred a younger age of disclosure. While the perceived effects of disclosure can become more positive over time [19], without knowing the exact length of time between disclosure and survey administration, this was not assessed in this study.

Conclusions

Disclosure of childhood genital surgery to women with CAH has increased over time. Although timing of disclosures varied, women with CAH and parents of daughters with CAH preferred disclosure, initiating it before age 14y and complete disclosure before age 18y. Disclosure may benefit relationships with parents and physicians.

Ethics approval

This study was approved by the Institutional Review Boards at the following institutions: Indiana University (IRB #1903250601), Children's Hospital of Los Angeles (IRB# CHLA-19-00463), and Weill Cornell (IRB# 1706018326).

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Conflict of interest

The authors have no conflicts of interest to disclose.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jpuro.2024.09.030>.