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Stigma Associated with Classical Congenital Adrenal Hyperplasia in Women’s Sexual Lives

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Abstract The risk of intersex-related stigma often serves as social indication for “corrective” genital surgery, but has not been comprehensively documented. In preparation for the development of an intersex-specific stigma assessment tool, this qualitative project aimed to explore stigma in girls and women with classical congenital adrenal hyperplasia (CAH) due to 21-hydroxylase deficiency. As part of a comprehensive follow-up project, 62 adult women with classical CAH (age range 18–51 years) took part in an open-ended retrospective interview focusing on the impact of CAH and its treatment on various aspects of girls’ and women’s lives. Deductive qualitative content analysis (Patton, 2014) of de-identified transcripts involved categorization of three types of stigma: experienced, anticipated, and internalized. Two-fifths of the participants reported CAH-related stigma in romantic/sexual situations. Stigma enactment by romantic partners occurred in reaction to both genital and non-genital sex-atypical features of CAH and sometimes included explicit questioning of the women’s true gender. Stigma anticipation by the women and their related avoidance of nudity, genital exposure, and romantic involvement altogether were frequent. Internalization of stigma occurred as well. In conclusion, the data suggest that many women with CAH experience, anticipate, and/or internalize intersex-related stigma in the context of their romantic/sexual lives.

Keywords Stigma · Disorders of sex development · Intersexuality · Congenital adrenal hyperplasia

Introduction

The birth of a child with genital ambiguity (aka intersexuality, now subsumed under the medical term, “disorders of sex development” [DSD; Hughes et al., 2006]) raises the question of appropriate gender assignment, which may cause parents much distress (Suorsa et al., 2015; Wisniewski, 2017; Wisniewski & Sandberg, 2015). The genital status is frequently believed to put the child at risk for social stigmatization (Oliveira, de Paiva-e-Silva, Guerra-Junior, & Maciel-Guerra, 2015; Rolston, Gardner, Vilain, & Sandberg, 2015). Early “corrective,” “feminizing” surgery of the genitalia—meant to “normalize” their appearance in line with the assigned gender—is often chosen as a clinical countermeasure to prevent or at least reduce intersex-related stigma (Feder, 2006; Hughes et al., 2006; Lewis, Ehrhardt, & Money, 1970; Meyer-Bahlburg, 2002; Money, Hampson, & Hampson, 1955; Mouriquand et al., 2016; Speiser et al., 2010). Because of inherent risks to cosmesis (appearance) and to function, both urinary and sexual, such surgery is controversial, especially when done primarily for social indications and before the patient has reached the age of consent (Binet, Lardy, Geslin, François-Fiquet, & Poli-Merol, 2016; Cools et al., 2016; Diamond & Garland, 2014; Frank, Mouriquand, Caldamone, & Malone, 2014; Lee & Houk, 2014; O’Connor, 2016).

Critics of early genital surgery for social indications rightfully point out that systematic documentation of both the stigmatizing effects of genital abnormality and the claimed social benefit of such surgery is lacking (e.g., Diamond & Garland, 2014; Lindemann, Feder, & Dreger, 2010). Indeed, most of the empirical evidence consists of scattered case notes and a few detailed case reports (e.g., Davis, 2015; Dreger, 1999; Karkazis, 2008; Money,

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1991; Preves, 2003). There are two exceptions. One is a Swedish study of a clinical sample of women with congenital adrenal hyperplasia (CAH) that included stigma in ratings of the impact of CAH, but no specific data on stigma types or social contexts (Frisén et al., 2009). The other is a U.S. study of parents' reports about their children (newborn to 17 years of age) on a written questionnaire comprising two scales, child-focused and parent-focused, and three subscales, perceived stigmatization, future worries, and feelings about the child's condition (Rolston et al., 2015); yet also in this study, the items are rather generic and do not provide specifics on aspects of the CAH condition that give rise to stigma or the related social contexts. At least part of the reason for the lack of systematic studies of intersex stigma is the absence of a comprehensive, standardized assessment tool for direct administration to patients with intersexuality.

When preparing for the development of a comprehensive assessment tool that is useful for both clinical screening and research purposes, an investigator needs to show that intersex stigma occurs frequently enough to justify the use of such a tool and to identify the features of the intersex condition that elicit stigma as well as the specific social contexts, in which stigma occurs. The present article describes findings from a formative, qualitative study with these goals. It was conducted in women with the most common form of intersexuality, namely classical CAH due to 21-hydroxylase deficiency (21-OH-D) in chromosomal (46,XX) females. This syndrome is associated with variable genital masculinization present at birth, which ranges from labial fusion and slight enlargement of the clitoris to the appearance of a normal male newborn with an empty scrotum and includes variable malformations of the vagina and urinary tract (New, Lekarev, Mancenido, Parsa, & Yuen, 2014).

Prior reports from this study focused on CAH-related stigma in the general social environment (Meyer-Bahlburg, Reyes-Portillo, Khuri, Ehrhardt, & New, 2017b) and in medical settings (Meyer-Bahlburg, Khuri, Reyes-Portillo, & New, 2017a). The present article addresses CAH-related stigma in the context of romance and sexuality, in which the genitals are likely to be particularly salient. As in the preceding two articles, the purpose of the current report is threefold: (1) to document the existence of context-specific intersex-related stigma (conceptualized as “undesired differentness” [Goffman, 1963, pp. 3–4]) and to highlight its emotional impact, (2) to identify plausibly contributing features of CAH, and (3) to make examples of stigma experiences readily available for use in the training of professional staff who care for such patients.

Method

Participants

The present study is part of a comprehensive project on the long-term outcome of 46,XX patients with classical or non-classical CAH due to 21-hydroxylase deficiency. As previously described

in detail (Meyer-Bahlburg, Dolezal, Baker, Ehrhardt, & New, 2006), the patients constituted a convenience sample recruited from among two subspecialty clinics in the northeastern U.S., but with residences distributed widely across the U.S. The present report is limited to patients with classical CAH living as women and includes 41 women with the more severe, salt wasting (SW) form of the illness, and 21 with the less severe, simple virilizing (SV) variant, all in the age range of 18–51 years (mean 30.54 years). Of the total of 62 women, 84% were white, 10% Hispanic, and 6% African-American. The average level of school completed was 5.25 on the respective 7-point Hollingshead scale (Hollingshead, 1975), in which “5” indicates partial college or specialized training. Almost all women with SW-CAH and more than half of those with SV-CAH had undergone at least one genital surgery on the clitoris and/or vagina, the first one mostly in early childhood. Some patients subsequently underwent further genital masculinization due to insufficient control of androgen levels, and many women underwent additional procedures in adolescence, most commonly to correct vaginal stenosis (stricture). All study procedures were approved by the respective Institutional Review Boards, and all participants provided written informed consent.

Measures

The data for this report were drawn from an open-ended, non-blindly administered interview that constituted the final procedure of an 8- to 10-h psychological evaluation battery of standard self-report questionnaires, psychometric tests, and (blindly administered) semi-structured interviews. The primary foci of the overall project were gender behavior and gender identity development from childhood through adulthood, sexual orientation, sexual behavior, and sexual dysfunction, and psychiatric disorders. The topical guide for the final interview focused on the impact of CAH and its treatment with hormones and genital surgery on diverse aspects of women's lives and was derived from two of the authors' (H. M.-B., A.A.E.) clinical experience with patients with intersexuality in general and CAH in particular. As described previously (Meyer-Bahlburg et al., 2017b), the questions were open-ended and did not explicitly use the terms “stigma” or “differentness,” but were worded so that reports of stigma-related experiences and feelings were facilitated. Early in the interview, the participants were asked: “In our experience, many children and teenagers with a medical condition harbor secret questions, expectations, hopes, fears and worries, and other thoughts about their medical condition and what it means for their future lives, that they sometimes do not even dare to share with anyone. When you think back to childhood, what secret hopes and worries did you have?” This was followed later in the interview using: “And at this stage in your life—what fears and worries, hopes, expectations, and questions do you have?” Probes focused on body image, gender role, dating/courtship, steady relationships, various aspects of sexuality including genital status and

sexual functioning, and parenthood. Interviews were conducted by women with M.A. or Ph.D. degrees in clinical psychology who were specifically trained for this project, except for a few instances, when scheduling problems made interviewing by the male lead author necessary. Depending on the time available and the degree of detail provided by participants, the interviews varied in length from about 0.5 to 1.5 h. The audiotapes of the interviews were transcribed by a transcription company certified in medical research confidentiality.

Data Analysis

To evaluate the applicability to somatic intersexuality of the three stigma categories used for chronic diseases by Earnshaw, Quinn, Kalichman, and Park (2013), we performed a qualitative deductive analysis (Patton, 2014, pp. 541–552) of the de-identified transcripts. Two of the authors (J. K. and H. M.-B.) excerpted women's statements (labeled "vignettes") that expressed CAH-related differentness, which in the given social context was perceived as adverse. Subsequently, at least two of the authors (J. K., J. R.-P., H. M.-B.) independently categorized these vignettes according to the three stigma types (experienced or enacted stigma; anticipated stigma including various forms of avoidance; internalized or self-stigma) and three categories of social context (social environment in general, e.g., family, peers; romantic/erotic situations; and medical settings). Disagreements regarding stigma types occurred mostly because many vignettes reflected more than one stigma type and some also more than one social context; they were resolved by permitting multiple codes of stigma type and context for a given vignette, and otherwise by clarifying the operationalization of categories (e.g., implying stigma anticipation in statements of stigma avoidance and categorizing general self-statements such as being "abnormal" or a "freak" as instances of stigma internalization). In addition, the diverse features of the CAH condition that gave rise to stigma were listed as well as the ways by which aversive differentness was signaled by others in the social environment and how the participants responded.

Results

Nearly two-thirds of the participating women with classical CAH provided accounts of stigma experiences (Meyer-Bahlburg et al., 2017b). The current paper provides data on stigma in the context of romantic and sexual partnering, as reported by two-fifths of the participants, primarily for late adolescence and adulthood. Examples are selected so as to highlight the diverse features of the CAH condition that gave rise to stigma and the ways by which aversive differentness was signaled by others in the environment. Stigma types are illustrated with examples that indicate the specific circumstances under which they occurred, the women's emotional responses and coping mechanisms, and some of the long-term sequelae in the women's romantic/sexual lives. Each vignette is

identified by the respective woman's code name (not her real name), followed by her age at interview, her CAH variant (SV or SW), and the vignette code (V #). Where helpful, the [bracketed] interview context or question posed by the interviewer (Int) precedes a woman's statement. Individual names included in some participants' statements were replaced with non-acronymic double-letter codes (e.g., "CC"). Some vignettes include more than one stigma type, but each vignette is only presented once to avoid repetition.

Experienced Stigma (Enacted by Sexual Partners)

When the women reported any reactions to their genitals by romantic or sexual partners, those partners' reactions typically focused on seen or felt genital atypicalities, which evoked intense adverse emotions in the women.

Megan (24 years, SV, V2): "There was a guy, CC, and ... like I did sleep with him. And he always wondered why I was always covering myself and stuff like that. And then one day, he just like flipped the lights on, and he was like, 'Oh my God! What's wrong with you?' and this and that. And that like freaked me out."

Even a just descriptive remark by her first partner caused the following woman to be vigilant with future partners.

Leonora (24 years, SW, V12): "There was one incident with my first partner. He had said something, and I ... I just played it off like I didn't hear him, so he wouldn't continue with it. But he did say something about my vaginal area. And right then and there I knew I had to be careful with, you know, who I was with and what to do with them, if they're mature enough to handle a situation like that. ... He made a comment about my vagina being flat."

Men may react strongly to noticing an enlarged clitoris in a woman.

Karen (32 years, SW, V16): [Partner's reaction to her genitalia:] "Oh God! What is it?" That's my rendition. I was more concerned about my clitoris and just that it was odd, in that sense. I was more concerned about the clitoris being hard, and, whenever I get nervous or whenever I clench down, which makes it move, and which makes me more nervous..."

One woman interpreted her enlarged and erectile clitoris as a "penis," i.e., a male organ, that was associated with unkind remarks by boys and undermined her self-image.

Linda (36 years, SV, V22): "When you have this penis and you go and you're trying to date a boy, how many boys do you know are not going to say some unkind remark about you having an erection? Oh, it's had a negative effect, of

course. I mean, having a miniature penis the majority of my life did absolutely nothing for my self-image.”

Also the male partners’ negative reactions to unusual genital features tended to include the explicit interpretation of these features as indications of maleness.

Megan (24 years, SV, V31): “If I get close with a guy, I never wanted to do anything with him, because I always thought I looked different down there, if he’d ask me like, ‘Hey, what is that?’, and this and that. Actually—it actually happened to me, where ... he saw me by mistake and was like, ‘Oh, you’re a guy!’”

Some men compared their woman partner’s erection to their own.

Hilda (23 years, SW, V26): [A man’s statement to her:] “‘Why does it feel as hard as mine?’ That’s the one [comment] that, like, got me ... I [also] get a lot of, ‘Are you a boy or a girl?’ I get a lot of that [maybe] because I was born with both genitals.”

Some women also reported men’s reactions to non-genital physical characteristics such as hirsutism or body build associated with CAH, which could raise gender doubts as well.

Linda (36 years, SV, V47): “One time ... I came out of a bar, and some guy was drunk, and he said, ‘Are you a man?’, and he started feeling my arms. And he goes, ‘Look how hairy your arms are!’”

Toni (32 years, SW, V50): “A lot of people, they also look at my features, they think I’m a guy, because they may think, ‘Oh shit, she’s not a girl, you’re a guy!’”

One woman found the gender-mislabeled so adverse that she limited her sex life to woman partners.

Dorothy (37 years, SW, V38): “DD [a man] who is about 22, 23, ... I started kind of wanting to become sexually active with him, and we were doing that petting thing, and he kind of asked me if I was a he-she, and that’s the last time I ever dated a boy [patient is now exclusively homosexual]. And even through my 20s ... I was probably on the extreme end of not sexually active. EE [another man] asked me if I was frigid, which I gave him. I just had no desire to let him see me or touch me in a physical way because of the CAH, the genital [issue].”

As one would expect, vaginal stenosis (stricture) and other problems of sexual functioning may complicate the situation further; such conditions led the following woman to distance herself from men and engage in alcohol abuse.

Stacey (38 years, SW, V40): “In my early 20s, I had some experiences probably with men, which ... I don’t know if

my vagina was too small and it was hard to have penetration ... and there wasn’t much lubrication, and so it wasn’t really a fun time. And I wasn’t having orgasms. So, yeah, there was a lot of [problems] ... And so ... I mean, stayed away from men and drank and, you know ... I wondered if you see any like alcoholism or drug addiction with people, with women with CAH.”

Anticipation/Avoidance of Stigma by Romantic or Sexual Partners

Women seemed to associate sexual situations with potential embarrassment and to resort to their avoidance.

Hilda (23 years, SW, V66): “It’s not like a problem like to date or anything. It’s just when it gets to the sexual part. That’s when I feel it’s a problem ... I always start doing stupid little things to just get on their nerves, and they just say, ‘Oh, I don’t want to be with you.’—‘Okay, bye.’ Because once you get intimate, it’s gotten into the sexual part, so I just don’t try to do none of that at all.”

Dorothy (37 years, SW, V62): “The genital area, as far as dating, if I, you know, would decide to become sexually active with somebody, it’s a huge issue ... It took a lot for me to get naked with somebody. It took a huge amount of trust. Kept me from sleeping around.”

The following woman just avoided foreplay but not necessarily coitus.

Pamela (26 years, SW, V106): [Impact on dating:] “I mean, just my [deep] voice and the uterine [the patient has had a uterine prolapse for several years]. That’s it. Those are the only two factors in my whole life that have affected my life or whatever. [Int: Effect of uterine prolapse:] It has nothing to do with sleeping with somebody. I mean, I’ve slept with people. ... [Inaudible portion] ... [Int: Well, you said now that ... foreplay is a problem because somebody will notice.] Uh huh. ... I mean I go as far as I want to. [Int: And then you cut at that point?] Uh huh.”

More commonly, stigma anticipation and avoidance extended to dating in general. The following report suggests that this may be understood as an effect of generalization.

Dorothy (37 years, SW, V58): “I think it [CAH] makes us very self-conscious, very almost afraid to talk about ourselves or look at ourselves. ... I don’t so much have it anymore, but I tell you, when I was dating, it was a huge issue. Huge issue for me. Acceptance, you know, acceptance by others.”

For some women the extension of avoidance to dating was specifically related to vaginal problems.

Stacey (25 years, SW, V56): “The vaginal problem totally killed it . . . You can’t really explain it without saying the word disease. Friendships are fine. But, you know, anything—dating, personal, intimate, yeah, same thing.”

For others, it was related more to hirsutism or to a combination of genital and hair-growth issues.

Melissa (18 years, SV, V48): [Avoiding dating because of hirsutism:] “What if I have more arm hair than he does?”

Cindy (44 years, SW, V36): “I’m, you know, sexually incapable to a vast degree. I’ve got some missing parts. I have hardly anything in the way of a vagina. But mostly, it’s been the facial hair. It’s a terribly inhibiting thing socially. [Interviewer: How does that interfere with your dating?] How would *you* like to be a bearded woman? . . . The older I get, the more socially inhibited I find I am: I just don’t want to be bothered with it. I’m tired of explaining and explaining why I’ve got facial hair and no vaginal opening.”

For still others, having the CAH condition overall and feeling awkward about its potential disclosure to others suppressed dating altogether.

Stacey (25 years, SW, V52): “I don’t want to date anybody, because I don’t know when I have to divulge this [the CAH condition].”

Karen (32 years, SW, V45): “I didn’t have any relationships in terms of dating anyone when I was younger. Therefore, I certainly didn’t have any sexual relationship at that point in time. I just felt awkward or whatever about the whole thing. And you know, even now, sexual-wise, it’s just not a huge part of my life.”

Some women described the psychological processes involved in dating avoidance, for instance becoming inhibited just when starting to feel attraction to someone or actively pushing potential partners away (see also V66 above).

Carol (20 years, SW, V28): “I’m sort of shy when it comes to courtship or whatever, having a boyfriend or whatever. It’s not easy for me to open up to a person, because . . . there are the questions of how different you are from other people, so a lot of things go through your mind. So that, I think, is what makes me a little more reserved in that sense . . . When it comes to trying to talk to somebody that I feel attracted to, I get shy.”

Kim (42 years, SV, V21): “What if I meet that guy, and it’s *the* guy, and I want to do something with him and can’t. Or . . . if he tries to put it in, and it doesn’t go in. I was kind of pushing people away because of that.”

Avoidance of sexual situations may also extend to potential women partners.

Dorothy (37 years, SW, V83): [Int: Impact of genital appearance:] “You know, I would never let anybody, when I first started to date, actually see it. . . . I kept that well discreet and hidden, you know, covered. . . . And . . . I really discouraged oral sex [by women partners] because of that reason - not about giving, but receiving, just because of the genital issues. . . . That has really taken a long time to get to that point [of permitting oral sex on herself]. . . . and I directly attribute that to learning more about the CAH, and I’m so disappointed in that it took me thirty-plus years to get there.” [Int: Anything else regarding barriers to nudity:] I’m sure that there’s been women I wanted to date that didn’t want me because my breasts were small, but nothing that anybody ever said to my face, you know. . . . There wasn’t a whole lot there to touch. They were just mere mosquito bites.”

Clitorectomy, as frequently practiced in the past, does demasculinize by removing the overgrown and sometimes penis-like clitoris, but may in fact overcorrect and, thereby, also leave a non-normal genital appearance that could make a woman avoid specific sexual practices with men.

Leonora (24 years, SW, V105): [Impact on sex with past partners, post-clitorectomy:] “I don’t want them to question me. So, I stayed in the dark. Not too much touching was going on. Because I figure, you touch, you—you ask questions. So, the only person who’s ever seen anything, besides the doctors, is my husband. . . . Sometimes, I even have problems with him touching me. But - [Int: Why?] Because there is nothing there.”

The following woman expected women partners to easily notice this post-surgical unusual appearance and, as a consequence, she kept away from sexual relations with women.

Leonora (24 years, SW, V104): [Impact on sexual imagery:] “There are certain things that I do want to experience, but I won’t because . . . of my shyness. [Int: Like?] I’ve—you know, had fantasies of being with the same sex. But, of course, that would be like the dumbest thing to do, because right off hand, they would know something is wrong. [Pt had had clitorectomy.] And, I’m not going to explain it to them, so I just leave it alone.”

Some women attributed their long-term single status to avoidance of sexual involvement.

Rosalyn (44 years, SV, V63): “I held back a lot . . . sexually . . . because I was feeling embarrassed. Maybe if I could be more forward, I would have been married by now.”

Internalization of Stigma

As a consequence of stigma experiences, the self-image of some women became one of being abnormal (see also V22

above). Stigma so internalized could add further to adverse consequences for dating and for future expectations of long-term partnership and marriage. One woman mentioned that the heritability of the CAH condition also added to her relationship barriers.

Anne (43 years, SW, V13): “I think [CAH] affected my interest in even bothering to date. So, that delayed everything ... I guess, you know, a feeling of kind of self-worth, you know, was lower ... I wasn’t interested. I enjoyed more kind of playing with the guys than dating the guys ... I always felt like I had a secret that couldn’t be told or discussed, or, you know, just a lack of openness I guess.”

Melissa (18 years, SV, V9): [Referring to her clitoromegaly:] “You know, my private abnormality. In the future, how do I go about, you know, when I do get married, or when I am in a relationship, how do I tell my partner that this is what I have to deal with. And there’s a chance that, you know, that my kids can get it if he’s a carrier. And do I really want to put that kind of burden on them. This is a disability, in a sense.”

The development of an abnormal self-image could also be facilitated by stigma coaching, for instance, in the context of CAH treatment.

Susan (51 years, SW, V5): “We’ve talked about how we try to pass for normal ... My main [worry] was that everything that I heard about my treatment was that this would make me be like other females. That I would grow up and I would marry and I would have children ... This did not resonate with me at any time in my life. So it simply emphasized to me that there was definitely something abnormal about me that I should never talk about ... and I just thought, well, that must mean there’s something wrong with me.”

Some women reported creative coping mechanisms, for instance, practicing some form of compensatory skill.

Jane (47 years, SV, V89): [Int: Effect of hirsutism on dating:] “... What you feel that is abnormal, if I have to say, you make up with something else. ... You draw away from that [abnormality]. But what you do is you make up with personality, either sense of humor, sarcasm, or both, which I have developed throughout the years. So, you’re not focusing on my looks or anything; you’re focusing on me, and essentially my mouth. ... You do develop a sharper tongue, though. Well, I have. I have. And I guess that’s just a defensive mechanism. Because what—what happens is—and again, this is, when you’re younger—you don’t know if you’re being stared at—So, like I said, you detract from that. And you—develop pluses in your personality, and then it becomes secondary.”

Genital Surgery as Stigma Relief

Given the women’s experiences of the association of genital atypicalities with stigma in its various forms, genital surgery could offer some relief, by resolving the sexual ambiguity of the genitalia.

Anne (43 years, SW, V100): [When asked how she felt about undergoing surgery:] “Well, not having a clitoris versus having a miniature penis, to me, is like a no-brainer in terms of knowing what one’s sex is. ... It would have been, I would think, virtually impossible to know [whether you were a boy or girl] until you had your period. And then, I think that you’d still question whether, you know, what kind of, I’d have to say, a freak of nature you are.”

Others emphasized how the normalization of their genital appearance reduced their inhibition and facilitated dating and relationship formation.

Heidi (20 years, SW, V102): “When I was younger ... well, before I had my surgery, I did feel different, because I wasn’t normal-looking, part of my body ... And there was a sense of [not] letting people get close to me. Like in the way of a relationship. ... I’m very shy ... I was just wanting to be in a group of people. [Interviewer: ... impact of surgery?] I guess it made me more open, meaning that if I was going to pursue a relationship, I knew that that wasn’t going to stop me from, you know ...”

Erica (26 years, SW, V101): [Prior to recent genital surgery:] “I never dated. Now, with the surgery ... I was afraid with the way the vagina looked, and so was always to myself. But now, since the surgery, I have more confidence ... It kept me in hiding for a long time. I wouldn’t do nothing before the surgery; now I probably will.”

Such relief could extend to non-sexual nudity situations as well.

Carol (20 years, SW, V103): [After describing her discomfort when putting on a bathing suit in the presence of other women before she had genital surgery:] “As for now [after genital surgery], I think, you know, I’m still a bit shy ... But, I think, you know, it’s better. I don’t—I don’t have a problem changing in front of my friends if I have to.”

Yet, for some, genital surgery itself may enhance the feeling of differentness, even though it may facilitate sexual intercourse (see also V105 above).

Maria (31 years, SV, V78): [Int: Impact of CAH:] “I didn’t feel normal when I was to go to have [genital] surgery ... when I was twelve and a half. ... I think that’s why, when I

was a teenager, I acted out and that's why I fooled around with people and because I wanted to be normal. . . . That's why I was promiscuous. . . . I thought that was the only way that I, you know, that I would be accepted. . . . That's the way boys would like me."

Linda (36 years, SV, V95): [Disclosure to others:] "You know, my husband's never made any comments about my clitoris or anything like that. But . . . He doesn't even know that I had surgery. I mean, I don't know if other women with CAH have divulged that information. . . . I—I'm just ashamed of it. 'Hey, YY, you know they hacked off my clitoris two times'. You know, it's—it's a shame, you know. . . . He knows it as adrenal insufficiency. He doesn't know it as CAH. . . . when you start throwing in CAH . . . , he can go on to the websites if he's having a boring night and start hitting all these websites, and there's all kinds of negative stuff out there about it." [This woman is particularly concerned about being considered half male, half female.]

Discussion

As the cited vignettes illustrate, many participating women with classical CAH experienced stigma ("aversive differentness") in the context of romantic and sexual situations, and this stigma was associated with the genital and non-genital physical features caused by CAH. All three types of stigma—enacted, anticipated, and internalized—that have been described for chronic diseases (Earnshaw et al., 2013) were also readily identified in our participants' reports.

Romantic/sexual partners' negative comments were elicited by both genital features such as overall appearance, clitoromegaly, and clitoral erections, and non-genital somatic symptoms of androgen excess, especially hirsutism. Both the women and their partners often appeared to interpret the noted atypical physical features of classical CAH primarily as atypicalities of sex, with some partners explicitly questioning the women's true gender and, thereby, possibly contributing to a perceived threat to the women's basic social identity (Major & O'Brien, 2005). Given the widespread stigmatization of homosexual and transgender individuals in the U.S. (Institute of Medicine, 2011), homophobia and/or transphobia may have contributed to the male partners' negative reactions to features suggesting physical maleness in women with CAH; exploration of this issue should be included in future studies.

The consequences of stigma for the women with CAH were manifold. According to their reports, stigma enacted by romantic/sexual partners led to immediate and strong emotional reactions such as distress, embarrassment, and shame. Such reactions to genitalia-associated stigma are probably facilitated by the early socialization of children in sexual modesty (Rosenfeld et al., 1984), i.e., to conceal their genitals from the view of others. In addition, many women had already become aware of undesired

differentness much earlier in development and were, therefore, sensitized to stigma long before they entered the adolescent and young-adult dating and sexual-initiation scene (Meyer-Bahlburg et al., 2017b).

As a way of coping with the risk of direct stigmatization, women with CAH resorted to hiding their body, and especially their genital area, from view and touch even during sexual encounters, adopted increased caution in sex-partner selection, and tended to keep their CAH status a secret. The related internalization of stigma is underlined by the low self-esteem and the self-labeling as "abnormal" that some women reported; whether this may also contribute to the increased rates of psychiatric problems such as depression found by some investigators (for references, see Meyer-Bahlburg, 2014) ought to be investigated in future studies. Longer-term coping behaviors included avoiding sexual encounters or abstaining from dating entirely, both of which interfere with partnership formation. Some women seemed discouraged from expecting any long-term partnerships such as marriage with a man in their future or gave up on romantic/sexual relationships with men altogether and adopted an exclusively lesbian lifestyle. One woman reported having resorted to alcohol abuse, a particularly damaging form of maladaptive coping. Many of the examples provided by the participants strongly suggest that CAH-related stigma and the heavy reliance on avoidance and related mechanisms of coping contribute to the well-documented delay of sexual milestones and increased rates of coital inexperience and of life without a sexual partner in women with CAH (for references, see Meyer-Bahlburg, 2014). Overall, the range of stigma effects and the women's diverse ways of coping are in line with findings by investigators studying other stigmatized (non-intersex) conditions (Earnshaw & Quinn, 2011; Hatzenbuehler, Phelan, & Link, 2013; Masnari et al., 2013). Reports of more positive coping mechanisms such as developing compensatory skills were rare, but this may have been a function of the focus of the interview.

A few women spontaneously reported on the expected or experienced beneficial effects of genital surgery, which is plausible if such surgery results in good cosmetic and functional outcome. Unfortunately, long-term follow-up studies of genital surgery in women with CAH have shown highly variable outcomes in both categories (Meyer-Bahlburg, 2014; Mouriquand et al., 2016). In fact, the majority of women in this study had undergone at least one genital surgery in their lives, and some of the striking examples of stigma provided above come from women who had had early genital surgery with unsatisfactory long-term cosmetic and/or functional outcomes. Improvement of surgical techniques in recent years has led to expectations for improved outcomes, but the confirmation of these claims rests on future follow-up studies of these younger cohorts when they have reached the age of regular sexual activity.

In the context of the controversy about early genital surgery, our findings suggest that stigma does play a much larger role in the lives of many women with CAH than recognized by the anti-surgery advocates. This fact by itself, however, does

not resolve the controversy. To support genital surgery—early or late—without reservations one would need to show that adverse side effects are indeed minimal, which is not supported by the data currently available. Concerning the timing of genital surgery, it needs to be established that early experience of genital stigma (i.e., prior to sexual initiation) is negligible or can be overcome by specific psychotherapeutic interventions, which currently also lacks satisfactory evidence. A recently published pilot study of deliberate delay of genital surgery to adolescence appears promising, given that “so far girls and their parents have not experienced significant concerns regarding genital ambiguity” (Bougnères, Bouvattier, Cartigny, & Michala, 2017), but the sample was very small ($n = 7$ girls with CAH), was young in age (1–8 years, median 4.5 years), possibly reflects self-selection bias on the part of parents who decide for surgery delay, and the study lacks systematic and detailed assessment of stigma and social context. Moreover, for some women, the mere fact that they had to undergo genital surgery is a reason for shame. Thus, even successful surgery does not appear to fully eliminate the need for some form of intersex-related psychosocial intervention for at least some women with classical CAH.

This study has several limitations. The sample was a convenience sample drawn from subspecialty clinics, and its representativeness for 46,XX women with CAH in general could not be ascertained. As most women had undergone at least one genital surgery, which is typical for the current generation of adults in the U.S., our sample does not include a subgroup of unoperated women covering the entire spectrum of genital masculinization that would permit a preliminary exploration of differences in stigmatization between operated and unoperated women. Ideally, stigma types and severity would also be correlated with the degree of deviation of genital appearance—e.g., clitoral size—from the norm (based on physical examination) at the time of the stigma experience, which in this retrospective study was not possible. Moreover, given the nature of qualitative interviews and the variable time constraints under which the final study interviews were conducted, women were not systematically asked about each potential facet of the entire stigma spectrum, which would require a comprehensive and detailed screening interview or questionnaire; even some of the probes were occasionally curtailed. Finally, those few women who—due to scheduling problems—were interviewed by a male interviewer may have felt more inhibited in disclosing their sexuality-related experiences.

Nevertheless, the findings of this study suggest that intersex-specific stigma of the three types, experienced, anticipated, and internalized, is quite common in the romantic/sexual sphere of life of women with CAH. The detailed material obtained in the overall project—on somatic features eliciting stigma, the social context in which it occurs, and the women’s reactions and coping responses—prepares the way to construct a comprehensive stigma screening tool specific to individuals with somatic intersexuality, which could be used for both research and clinical purposes.

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