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Meyer-Bahlburg, Heino F.L.; Reyes-Portillo, Jazmin; Khuri, Jananne; Ehrhardt, Anke A.; and New, Maria I., "Syndrome-Related Stigma in the General Social Environment as Reported by Women with Classical Congenital Adrenal Hyperplasia" (2017). *Department of Psychology Faculty Scholarship and Creative Works*. 470.

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Syndrome-Related Stigma in the General Social Environment as Reported by Women with Classical Congenital Adrenal Hyperplasia

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Received: 26 November 2014 / Revised: 2 September 2016 / Accepted: 8 September 2016 / Published online: 27 September 2016
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Abstract Stigma defined as “undesired differentness” (Goffman, 1963) and subtyped as “experienced” or “enacted,” “anticipated,” and “internalized” has been documented for patients with diverse chronic diseases. However, no systematic data exist on the association of stigma with somatic intersexuality. The current report concerns women with classical congenital adrenal hyperplasia (CAH), the most prevalent intersex syndrome, and provides descriptive data on CAH-related stigma as experienced in the general social environment (excluding medical settings and romantic/sexual partners) during childhood, adolescence, and adulthood. A total of 62 adult women with classical CAH [41 with the salt-wasting (SW) variant and 21 with the simple-virilizing (SV) variant] underwent a qualitative retrospective interview, which focused on the impact of CAH and its medical treatment on many aspects of women’s lives. Deductive content analysis was performed on the transcribed texts. The women’s accounts of CAH-related stigma were identified and excerpted as vignettes, and the vignettes categorized according to social context, stigma type, and the associated features of the CAH condition. Nearly two-thirds of women with either variant of CAH provided stigma vignettes. The vignettes included all three stigma types, and most involved some somatic or behavioral feature related to sex or gender. Stigma situations were reported for all ages and all social contexts of everyday life: family, peers, colleagues at work, strangers, and the media. We conclude that there is a need for systematic documentation of stigma in intersexuality as a basis

for the development of improved approaches to prevention and intervention.

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

Introduction

Individuals with intersexuality, in current medical terminology subsumed under the broader category, “disorders of sex development” (DSD; Hughes et al., 2006), usually develop a sex-atypical reproductive tract, variably associated with sexual ambiguity of the external genitalia, secondary sex characteristics, body build, and voice. Appearing different in physique or behavior than expected is likely to draw the attention of others in the social environment, especially if the differentness is valued negatively. “Undesired differentness” associated with being denigrated by others is a widely used definition of stigma (Goffman, 1963, pp. 3–4). Stigma theorists differentiate several subcategories of stigma (Link & Phelan, 2001). As the concept has been applied to patients with diverse chronic diseases (Earnshaw, Quinn, Kalichman, & Park, 2013), in this report we distinguish between experienced stigma, i.e., being actually exposed to stigma enacted by others; anticipated stigma, i.e., expecting exposure to stigma to occur in specific situations (commonly associated with attempts to avoid those); and internalized stigma or self-stigma, i.e., seeing oneself as abnormal or otherwise denigrating oneself. Self-stigma may be strengthened by “stigma coaching,” i.e., efforts by others to protect a vulnerable individual from stigmatization, but thereby unintentionally corroborating that the stigma-eliciting feature really is “bad” (Schneider & Conrad, 1980). Chronic diseases and other conditions vary in the extent to which they are noticeable or concealable (Quinn & Chaudoir, 2009), which in turn may affect the degree of stigmatization. For people with chronic diseases as a

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group, stigma has been shown to have an adverse impact on quality of life and even on health care (Earnshaw & Quinn, 2011; Earnshaw, Quinn, & Park, 2011).

The extent to which somatic intersexuality is associated with stigma is a matter of controversy. Many intersex-experienced clinicians are aware of individual patients and families in whose lives intersex-related stigma plays or has played a significant role, as was already recognized in the early psychological studies on CAH (Ehrhardt, Evers, & Money, 1968). Such cases serve as justification for certain policies of clinical management, such as early “gender-confirming” or “normalizing” genital surgery to bring the genital appearance in line with the assigned gender (Lewis, Ehrhardt, & Money, 1970; Meyer-Bahlburg, 2002; Money, Hampson, & Hampson, 1955; Speiser et al., 2010) or the cautious disclosure of details of a patient’s condition and medical history to people in the patient’s family and social environment and, later, the deliberately gradual disclosure to the patient him- or herself (Hughes et al., 2006; Meyer-Bahlburg, 2008; Money, 1994). Many intersex activists, however, oppose such policies of clinical management, especially genital surgery, as being more done for the comfort of the parents or in support of the gender ideology of society than for the good of the patient (e.g., Chase, 1998, 1999, 2003). Yet, in this type of discussion, the actual stigma experiences of people with intersexuality are usually only illustrated by a few individual anecdotes. Also, recent critics of gender-confirming genital surgery point to the lack of systematic data on intersex-associated stigma (Diamond & Garland, 2014; Lindemann, Feder, & Dreger, 2010). The published documentation of intersex-related stigma is mostly limited to brief case notes (e.g., Karkazis, 2008; Preves, 2003) and a few detailed case reports (e.g., Davis, 2015; Money, 1991), except for one study of a clinical sample that included stigma in ratings of the impact of the patients’ intersex syndromes (Frisén et al., 2009). Systematic stigma research in this area is held back in part by the logistical problems associated with the low prevalence of the many and diverse intersex syndromes as well as by the lack of standardized methods for the assessment of intersex-specific stigma.

As a first step toward the development of systematic methods of assessment and intervention, our team has conducted qualitative studies of stigma in the most prevalent intersex syndrome, classical congenital adrenal hyperplasia (CAH) in chromosomal (46, XX) females. In this syndrome, a genetically caused enzyme defect variably affects adrenal steroid production leading to overproduction of adrenal androgens, which produce variable degrees of prenatal masculinization of the genitalia and postnatal virilization of the body associated with additional risks to health (Acherman & Hughes, 2011; New, Lekarev, Mancenido, Parsa, & Yuen, 2014). CAH also affects diverse categories of long-term psychological outcome (Meyer-Bahlburg, 2014). The syndrome requires life-long hormone treatment to replace the underproduced cortisol and, in the salt-wasting (SW) variant, also aldosterone. As intersex-related stigma is a potentially highly sensitive area of inquiry, we decided to conduct a first such project not with children and

adolescents, but retrospectively with adults. A first publication from this study concerned CAH-related stigma experiences in medical settings (Meyer-Bahlburg, Khuri, Reyes-Portillo, & New, 2016). The current report covers CAH-related stigma experiences in the family, with peers, and others in the general social environment, and stigma internalization during childhood, adolescence, and adulthood. As in the preceding publication, the purpose of the current report was threefold: (1) to document the existence of intersex-related stigma (Goffman’s “undesired differentness”) and to highlight its emotional impact, (2) to identify plausibly contributing factors, 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 current study is part of a larger project on the long-term outcome of adult women with either classical or non-classical CAH due to 21-hydroxylase deficiency who were recruited from one of two subspecialty clinics in the Northeastern USA as described previously (e.g., Meyer-Bahlburg, Dolezal, Baker, Ehrhardt, & New, 2006; Meyer-Bahlburg, Dolezal, Baker, & New, 2008). All study procedures were approved by the appropriate institutional review boards, and all participants provided written informed consent. The final sample for the current study included 62 women with classical CAH: 41 women with the more severe, SW variant and 21 with the less severe, simple-virilizing (SV) variant. Participants ranged in age from 18 to 51 years (*M*, 30.5 years); 84 % were White, 10 % Hispanic, and 6 % African-American. 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, but some have had subsequent further genital masculinization due to insufficient control of androgen levels.

Procedure and Measures

As outlined previously (Meyer-Bahlburg et al., 2016), all women underwent an 8–10 h comprehensive protocol including various questionnaires and interviews assessing the long-term outcome of CAH. The protocol ended with an open-ended, nonblindly administered interview, which focused on the impact of CAH and its medical treatment on diverse aspects of women’s lives. This final interview provided the data for the current study. The topical guide for this interview was based on two of the authors’ (H. M.-B., A. A. E.) clinical experience with patients with DSD in general and CAH in particular. It was designed to elicit accounts of the women’s experiences of the diverse aspects of the CAH condition and its treatment with hormones and surgery at different ages from childhood through adulthood and in diverse social environments.

The questions were open-ended and did not explicitly focus on “stigma” or “differentness,” but were worded so that reports of stigma-related experiences and feelings were facilitated. For example: “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?” Then later: “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, and parenthood. Interviewers were M.A.- or Ph.D.-level clinical psychologists specifically trained for this project. Almost all interviews were conducted by women. Interview length varied from about 0.5–1.5 h, depending on the degree of detail participants provided and the time available.

Data Analysis

All interviews were audiotaped and transcribed by a transcription company certified in medical confidentiality. Subsequently, a deductive content analysis was performed (Patton, 2014, p 541–552), based on the conceptualization of stigma in the context of chronic diseases by Earnshaw et al. (2013). In the de-identified transcripts, the women’s accounts of experiences that reflected “undesired differentness” or stigma were identified and excerpted as vignettes by two of the authors (J. K. and H. M.-B.). Each vignette was independently categorized by at least two of the authors (J. K., J. R.-P., H. M.-B.) according to the three context categories (social environment in general, e.g., family, peers; romantic/erotic situations; and medical settings) and as to stigma type (experienced or enacted stigma; anticipated stigma, here usually implied by various forms of avoidance; internalized or self-stigma, here mostly indicated by labeling oneself abnormal). Disagreements initially occurred mainly due to the fact that many vignettes reflected more than one stigma type and some also more than one context. They were resolved by permitting multiple codes, and otherwise by clarifying the operationalization of the categories. In addition, the diverse features of the CAH condition that gave rise to stigma were listed as well as the ways by which undesired differentness was signaled by others in the social environment and how the participants responded. The current report focuses on stigma in the general social environment.

Results

Nearly two-thirds of women with CAH reported stigma experiences (15/21 SV; 23/41 SW). In the text below, we illustrate the three stigma types by age group. Examples are selected so as to highlight the diverse features of the CAH condition that gave rise to stigma, the ways by which adverse differentness was signaled

by others in the social environment, and some of the women’s emotional responses and coping mechanisms. Each vignette is identified by the respective woman’s code 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 letter codes (e.g., “BB”). Although some vignettes include more than one stigma type, each vignette is only presented once to avoid repetition.

Stigma Conveyed by Parents and Media or Enacted by Peers in Childhood and Adolescence

Stigma Conveyed by Parents and Media

Direct enactment of stigma by parents was extremely rare. However, some parents conveyed adverse differentness by conspicuous silence, especially in regard to a girl’s genitals. While scanning vignettes for examples of adverse differentness, we came across several descriptions of situations, in which young girls with CAH became aware that their genitals looked different from those of other girls, but without obvious connotations of adverseness. For some girls with CAH, bathing with another girl was the occasion when they first noticed a genital difference.

Dorothy (37 years, SW, V73): I had no clue that I was different until me and [girl BB]...saying I let you look, if I can look...I believe we were like 10 or 11.

Publications dealing with sexual anatomy provided other opportunities for genital comparisons.

Linda (36 years, SV, V80): We had a bathroom...and in the bathroom cabinet, there was pornography magazines. And I remember this to this day. So I knew as a young child what a woman’s vagina is supposed to look like, and I knew I looked different.

Whether the discovery of having genitals different from those of other girls or women became an adverse experience appeared to depend in part on parental reactions. Several parents conveyed via silence and secretiveness that having atypical genitalia is a stigmatized condition.

Susan (51 years, SW, V68): Learning that you don’t talk about this—this kind of hit home with me when I was about 7, and it was right after—we had just our baths—my sister, AA, and I...we were examining each other. I was counting...and I said, “Mommy, AA has three holes.” And AA is counting me, and she could find only two. And I just thought this was wild. I only had two holes that AA could find. And Mom just looked at me and said, “Get to bed.” And just—you know, usually, Mom is like—I’ll ask why the sky is blue, and she’ll give me this long thing about refraction and dust in the

atmosphere. That question—“Get to bed”—and I knew that was a topic we don’t discuss.

Some parents discouraged genital exposure through sex play.

Eve (38 years, SW, V72): [Int: Impact of having CAH on sex play in childhood:] I remember when Mom and Dad would have, you know, sit us down and say, “Now, guys...you can’t be doing this,” you know. And it seemed to be much more of an issue for my brother who’s affected [with CAH] and my sister and I [both affected] than for my other [unaffected] three sisters.

Parents’ attempts to prevent genital exposure without providing explanation extended to other situations as well.

Tonya (31 years, SV, V33): My parents would never talk about it. As a little girl, I used to bend over in the yard to do things, and they would scream at me, because I could be exposed [i.e., her atypical genitalia]...And also, you know, little children like to walk around with just a T-shirt on, and they would never tell me the reason why they were asking me not to. But they didn’t want me to go play at friends’ houses because that’s the reason why they were scared.

Some vignettes indicated that the parents’ noticeable avoidance of all talk about genital matters combined with the girl’s sensitization by the frequent genital exams in her doctor’s office to enhance the stigma effect.

Susan (51 years, SW, V8): [Int: Re avoidance of romance/sex:] Well, the genital exams obviously have played a big part. It’s the whole thing. “We [parents] must never talk...we don’t discuss this.” Anything that has to do with the genitals just was never talked about. And then, of course, the examinations themselves...So, a combination of all this.

One woman remembered expressing to her mother how in her mind noticing genital differences from other girls combined with aspects of CAH treatment to underline her differentness.

Megan (24 years, SV, V81): Like when my mom used to put me and my cousin in the bathtub, I used to say, “Why I didn’t look like her down there and why do I always have to...get needles and take medicine,” and saying like, “I am different.”

For some girls with CAH, magazine articles about intersexed genitalia introduced doubts about being a girl. In the following example, explicit discussion with the mother (rather than parental silence) helped to alleviate the girl’s doubts about her gender.

Melissa (18 years, SV, V93): I used to think that I was half boy, half girl, for some reason...[Int: Origin of that notion?] I read also, maybe in *Newsweek* or *Time*, about children that are, you know, they have both sexes in them, and you can tell by their private parts. And my clitoris isn’t that large to, you

know, assume that. [Int: From which age on?] Maybe fourth grade, fifth grade on. [Int: Till when?] Maybe till I really started to hit puberty and kind of go through...you know, the big change. [Int: How did that get resolved?] Yeah, well, it got resolved when...I talked to my mom about it as well, and she tried to explain her best. And I took another look at that booklet [(about CAH)]...And I examined it again...because they always say that it [genitals] may look male-like. So that’s...what I think really spurred that image into my head. But...but I have overcome it. There’s nothing male-like about me.

Surgery to correct marked masculinization of the genitalia sometimes may have unexpected effects in terms of a girl’s perception of her genital differentness.

Leonora (24 years, SW, V82): [Int: Impact of having CAH on sex play in childhood:] Yeah. I didn’t...I was...at the time [after clitorectomy at age six years], I was ashamed... Because, I mean, they’re little boys, but they know what they’re looking at, and...so, I didn’t participate in those kind of games.

Our interviews yielded only one example of direct stigmatization by a parent.

Linda (36 years, SV, V6): My mother didn’t even want to have anything to do with my [medical care]. My mother used to call me and my sister [also CAH-affected] “medical rejects” when we were kids.

Stigma Enacted by Peers and Others

Experiences of stigma expressed or enacted by peers were reported already for childhood, but more frequently for adolescence.

Differentness could be experienced by the mere fact of having to take medications.

Simone (20 years, SW, V42): I always kind of felt like an outsider...When I was younger, you know, go to a birthday party and just...one kid would ask me about medication and everybody starts asking why.

Some women mentioned discomfort about disclosing their CAH-related doctor visits to peers.

Karen (32 years, SW, V46): I took several trips a year to [city with medical center] as a child and had to take time out of school as a kid, had to explain or try not to explain why I was leaving to my friends and that kind of thing. For some reason, that kind of thing was definitely in my life and causing me angst of some sort...I was a pretty fearful kid in terms of my fear with not wanting to die, and that...definitely popped its ugly head up when I was a child quite often.

Occasionally, a CAH-related behavior would result in some teasing or a practical joke as in the following illustration from a woman with CAH who compensated for salt loss by increased salt consumption.

Susan (51 years, SW, V96): There was a problem for a while there on the salt thing. I remember I was sort of teased because...I remember one time when it was...oh, gosh, about the second grade, and I went to get something, and I came back and I started drinking my milk, and it was loaded with salt. And, you know, this girl, who was supposedly my friend—this was a practical joke—she thought this was hilarious. And I—I was just so bitter and angry at her for doing it.

During adolescence, differentness—related strongly to atypical pubertal maturation—was conveyed by peers' stares, comments, and questions. Hirsutism was a particularly frequent issue.

Nina (28 years, SV, V61): The body hair's definitely been an issue. You're always wondering, "Why are they staring at me?"

Barbara (22 years, SW, V64): I got teased by it. Oh, just that, you know, they would comment. People would comment about my...more or less about my arm hair and say about how, you know, I'm hairy.

Hirsutism and other sex-related physical features made peers question a girl's gender, which was particularly hurtful.

Rebecca (23 years, SW, V49): [People said:] "You look like a boy." Or, "your haircut, your face"...I cry sometimes. When I was younger, I would cry. I just don't feel right to be called a boy. I don't think boys would want to be called girls, either.

Johanna (51 years, SV, V55): When I was in junior high school, you know, kids would stop me and look at me like, you know, I was a freak, and I remember one girl said, quote, "Maybe you were meant to be a man." And that was extremely, extremely upsetting. I'll never forget that. I've even had people talk—in the building—talk about it, make an issue out of it. A lot of people—of course junior high school kids, junior high school age, you know, are very cruel—but even adults would make comments about it [severe facial hirsutism].

Also the delayed or diminished growth of breasts elicited teasing by peers.

Tonya (31 years, SV, V27): [Int: barriers to getting sexually involved:] With no breast development, of course, you know, I didn't feel attractive at all...I think the breast thing came along because I was teased a lot about that part. Being in high school, it's so big, and all the other girls [were developing]...I was teased a lot by my peers about that. I

think that's what's...locked into my mind that that was the really important thing.

Because of their different physical appearance, some girls with CAH experienced exclusion from the peer group.

Linda (36 years, SV, V51): [In regard to acceptance by the peer group:] You know, if you look a little masculine, and you have all these prissy girls, and you know, they don't want you to be part of their group because you don't look like them...

Stigma Enacted by Others in Adulthood

In adulthood, many women with CAH continued to experience stigmatization elicited by atypical body features. Hirsutism continued to be an issue.

Cindy (44 years, SW, V65): [Regarding facial hirsutism:] You are the subject of stares. Little children look and say, "Hmm, is that person a man or a woman, Mommy?" I've had kids come up and ask.

A new salient feature, not mentioned for adolescence, was a deep voice, which also made others question a woman's gender.

Linda (36 years, SV, V43): [Regarding acceptance by peers:] Like a lot of people get put off by a deep voice. You know, that's what I encounter as an adult. I've got a deep voice...It's deeper than average, I know that. And, you know, I worked with [a woman] last year where she made fun of my voice...So, yeah, I think it can affect your relationships.

Gender-atypical features sometimes combined in raising gender doubts.

Rosalyn (44 years, SV, V76): [Int: Impact of CAH on your life:] Only the hair growth and, you know, the voice. [Int: How?] Oh, just people say, "It's a man," you know...and stuff like that. It's just a little embarrassing things. It makes me more inhibited, I guess you'd say...And I felt bad that I couldn't have children.

Also in adults, being mistaken for a man in superficial encounters may elicit a stress reaction, although for this age group of less intensity than some women reported for the adolescent years.

Rosalyn (44 years, SV, V86): Oh yeah...I mean, today, like, even walking in here [hospital], then the escorts say, "I'll take him back." You know what I mean?...They're not doing it maliciously, and I'm not saying it is, you know. It's just I get a little red neck, you know...

Some adult women adopted a matter of fact way to correct such misidentification by others.

Elisabeth (30 years, SV, V59): [Regarding being addressed as “Sir” in public places:] I just try to correct them, tell them, “Ma’am”, and just go about my business.

Identification badges could be useful.

Erica (26 years, SW, V53): [Int: In regard to being mistaken for a man:] [People are] generally, “Sir, can I help you?” So, people used to call me a man, so now they still do... till they look at my work badge and it says my name on it... I look similar to a man. The body. My voice is deep.

Occasionally, the categorization of intersex in the media had a profound adverse effect even on an adult woman with CAH, as illustrated by this internet example.

Linda (36 years, SV, V97): I went on that Intersex Society site, you know... And when I first saw it, it really freaked me out. Really, truly did. Even as being someone 35 years old—I think that’s when I stumbled upon it last year. Do you know it really made me self-conscious of who I am... Because it’s kind of like you’re not either male or female there, you know... And to me, I consider myself a female. So their idea, because I have congenital adrenal hyperplasia, I’m kind of not? You know, so—oh, please, I could go on and on and on about that one.

In one vignette, being identified as a woman with CAH made that woman an object of unwelcome sexual curiosity.

Linda (36 years, SV, V69): [Int: Impact of CAH on a person’s life:] ... I tried being a pen pal with a CAH woman, and you know what she said to me? She said she’s always wondered what it was like to have sex with another woman with CAH. And I’m like, you know, you can’t even like—say—you’re in correspondence with someone without somebody getting these frigging stupid ideas, you know—[laughs]—and that was the last time I tried, you know, having a pen pal. It was like, well, that’s not going to work.

Anticipated Stigma and Coping in Adolescence

Stigma was often anticipated or “felt” (Link & Phelan, 2001), that is, girls or women with CAH were self-conscious of being different and anticipated stigmatizing reactions by others to visible physical characteristics.

Johanna (51 years, SV, V30): I had a lot of fears... Well, I mean, growing up with, you know, hair on your face and everything, it was just—I was just always fearful of what people would say about you. People always said something. Always. And very cruel. Oh, unbelievably cruel. [Int: In regard to CAH effect on body image:] Oh, tremendous. Tremendous. Of course, the hair on my face, the puffiness, what I was going through with my weight. Puffy, like a balloon.

Some women tried to cope by minimizing the chances that others could see the physical effects of CAH.

Rosalyn (44 years, SV, V37): [Regarding hirsutism:] I’ve been very self-conscious of it... I won’t get changed in front of people. Like in locker rooms, I was the fastest dresser. I’d have to be really comfortable with someone before I let them see it.

Changing in front of other young women could be a particularly stressful challenge for one woman with uncorrected clitoromegaly.

Carol (20 years, SW, V85): [Regarding barriers to nudity:] A typical example—you go for a swimming lesson or whatever and you have to change in front of all the girls. That was—that was very hard for me... Because I knew I was, you know, different from the other girls. And even putting on a bathing suit, you know, what happens if you think of something erotic, and then you have an erection. And, you know, at that time [before surgery], you know, it did make a difference for me. It showed physically, whether I did or not.

Even non-obvious features such as delayed menarche could affect participation in peer- group activities.

Deborah (40 years, SV, V23): [Regarding adolescent peer relationships:] The only impact that I remember being conscious that I wasn’t getting my period when other girls were. It made me stay out of certain discussions, because—I didn’t want to participate in them. I didn’t want people to know that I didn’t get my period yet.

The genital aspects of classical CAH constituted a particularly sensitive topic that needed to be kept from the peer group.

Susan (51 years, SW, V90): [Int: Impact of CAH on peer relations:] Well, as I say, a lot. I’ve devoted a lot of energy to hiding the fact that I had CAH. Talking about—the salt thing I could never hide. I mean a lot of people would ask me and I’d talk about it. And then they’d say, “Well, how did... they find out?” I never would ever mention that I had—that it was genitalia. It was always that I was vomiting because of—I was a salt loser...

For some, a need to avoid disclosure of CAH altogether profoundly interfered with forming close personal friendships.

Karen (32 years, SW, V19): In terms of close personal friends, I guess I really didn’t have a lot of that kind of friendships. In a way, I guess CAH kind of created in me a wall, and I guess I kept it up through everything, in terms of I didn’t confide in my friends my personal deep feelings or anything like that. We did things, but, yeah, they never knew I had CAH, and I never shared any emotional things with them.

Anticipated Stigma and Coping in Adulthood

Felt stigma or anticipated stigma also continued into adulthood. One woman described using her arms to conceal her limited breast development.

Dorothy (37 years, SW, V11): You know, the breast thing, when I was a teenager, and even probably through my 20s, was a huge issue for me. Huge to the point where I was always with my arms crossed over my chest. Sometimes I still am, but very, very rare... I do kind of look kind of goofy when I'm nude, and I'm not your typical female shape, in the hips, and my breasts are small. I have no hips. My body shape is, you know, really masculine, and I do see that.

Wanting to hide features such as hirsutism kept some women from participating in common activities with their peers.

Elisabeth (30 years, SV, V17): I've never been in a swimming pool with, you know, any of the other girls... and my girlfriend says, you know, "Don't worry about it," you know, because, you know, she was going to shave my back and everything so I'd feel comfortable around the other girls... A lot of people, that's why they mistake me for a man, because of the facial hair. You know, I have to shave. And the baldness.

A woman who was fond of rough activities such as trail building in the wilderness avoided such group activities, because she could not conceal her hair growth.

Cindy (44 years, SW, V15): I'm a member of [several wilderness societies]. They have all these wonderful programs that you can go out, can help restore trails, build trails, do these things. I can't go out in the damn wilderness! There ain't no electrical outlet on a tree. How am I going to cut... get the facial hair? You become a freak. You become a social freak within two to three days. You're up shit's creek. You talk about being apart... Who cares? They can't see what's underneath my underpants, but you sure can see what's growing on your chin and your lip.

Some young adults refrained from disclosing having "CAH" even to their close friends.

Melissa (18 years, SV, V84): [Int: Disclosure to women friends:] ...No one knows about my CAH. What my closest friends do know is that I do have a problem with my adrenal glands... But I don't go into specifics. There's just some things, you know, you need to keep—for me, I think, I need to keep some private things private.

Internalized Stigma in Childhood and Adolescence

Having to take medications daily conveyed a sense of not being normal.

Leonora (24 years, SW, V71): I knew that I would be on medication for the rest of my life. So, when I was younger, I would just think I was different and wanted to be a normal child.

For some girls, the need for hormone replacement was associated with the sense of being severely ill and the awareness of an increased risk of mortality.

Melissa (18 years, SV, V74): I was missing something, and I always felt if I don't take it [the medication] I could die... when I was younger.

Hilda (23 years, SW, V77): [Int: Secret hopes and worries when a child:] The only thing was like I thought I was like real, real sick, so I always like had like "How long do I have to live?"

Some girls developed a poor body image, due to obesity, for instance (although in this example, the woman adjusted to being overweight in adulthood).

Naomi (20 years, SV, V94): [Int: Impact of CAH and its treatment:] When I was younger, it was very hard for me to deal with it... I was heavy... I wasn't very confident when I was younger and always felt that I wasn't perfect enough for my age, looking the way that I did. But now—I'm at the point where I just don't care what everybody thinks anymore about me physically, because I know, you know, that I'm okay with it.

Even delayed menarche could make a young woman feel being abnormal.

Maria (31 years, SV, V88): [Int: Impact of CAH:] [Not having] periods... I didn't feel like a normal teenager.—I really always had friends. There was nothing ever an issue that would affect my social life. It was just my—inside me and how I felt about myself.

In some families, keeping CAH a secret and creating a façade of normalcy was the habitual coping mechanism.

Susan (51 years, SW, V79): [Regarding secrecy:] There was this—see, "you don't talk", but it was also this—"we all need to appear normal." This was such an important thing, that we do not—stand out as being different from others. And apparently I was succeeding at it, to even fool my sister who had CAH like I did, which amazed me that she was not able to see through that façade.

Internalized Stigma in Adulthood

Several women with CAH reported internalized stigma also for adulthood, with a strong focus on sex-atypical body characteristics. Even hirsutism alone sometimes undermined a good sense of oneself.

Elisabeth (30 years, SV, V91): [Int: How feminine are you:] I'd say about, I don't know—I'd say about 65 %... I feel like a lady and everything. It's just the hair growth on my back and the—having to shave and all that. If it wasn't for that, I think I would feel, you know, a lot more—a lot better about myself.

More commonly, multiple sex-atypical body characteristics combined for an undesired masculine body image.

Hilda (23 years, SW, V87): [Int: Impact on body image:] Like I'm too big... My arms, they are big. Like they're muscular. I feel too masculine. That's how I feel. [Int: And you attribute that to CAH?] Well, not really CAH, but the steroids I was on from a young age... And at the same time when I was taking it, it's like, I was like stronger than any girl.

Having masculine features conveyed a sense of not being normal-feminine.

Anne (43 years, SW, V67): [Regarding CAH effects on body image:] Sure... You know, I have a deeper voice. I'm short. And, you know, some of my features are more masculine than normal. Just in general. Facial features, my muscles.

For some women, the genital masculinization contributed further to the sense of not being a normal woman.

Cheryl (22 years, SW, V7): I kind of wished that I would just, you know, be normal like everybody else—just the genitalia issue of it... Like my mom always said, it's symptoms, I think, the masculinism of this—disease has kind of shown. Like, for instance, like my ankles, they're a bit larger than a regular girl's ankles would be. And like your wrists and stuff. Men can usually have a wider wrist or ankle than a woman does.

For one woman, the memories of stigma in younger years had an emotional intensity reminiscent of post-traumatic stress-disorder. The aftereffects of these memories became the basis for hypervigilance to potential stigma experiences of her own child and related protectiveness.

Tonya (31 years, SV, V1): [CAH] has left a lasting imprint in my mind and in my life that I think will never go away... All the emotional stress that I had way back, so that even talking about it even today, tears still come to my eyes. It has left a scar in my life that nothing—I don't think anything will remove it, even though life is a lot better [after surgery, patient now has a vaginal opening and reduced-size clitoris]. And the way I was treated because of CAH... I felt like someone coming from Mars, like an alien. I was always treated like someone who came from another planet. [I'm] different in the sense of now that I have a [biological] child, I will make it my duty that no one take

advantage of her, in the sense of tease her, no matter for what. It wouldn't be tolerated by me. Because I don't want her to feel the pain and have a scar left in her mind.

Discussion

The data clearly indicate that stigma defined as “undesired differentness” was quite common among girls and women with CAH. It appears likely that accounts of stigma would have been elicited from even more women if we had been able to use a structured interview that systematically covered the diverse aspects of stigma revealed in this qualitative study. Stigma was experienced by these women in all three forms: enacted, anticipated (as implied by avoidance), and internalized. However, in contrast to the ease with which one can formulate questionnaire items that distinctly represent the three stigma types (Earnshaw et al., 2013), there appears to be considerable overlap in real life as represented by the vignettes. Experience of stigma enactment by others may lead a woman to anticipate/avoid similar situations and may also be understood by her as an indication of being “not normal,” and whether one, two, or three of these categories of stigma are expressed in an individual vignette depended to some extent on the interview context.

Stigma-triggering situations were described for all ages and all social contexts of everyday life: family, playmates, schoolmates, colleagues at work, neighbors, and strangers. The sense of being undesirably different did not solely depend on direct pejorative comments by people in the social environment. It was also signaled by other people's mere attention to, and questions about, one's body characteristics and other aspects of CAH. Moreover, it could be elicited by intersex coverage in the media and even by the wording of educational materials designed for such patients and their families. Direct stigmatization by parents was rare in our sample, but the silence and secretiveness maintained by some parents around salient issues of CAH-related appearance and examinations seemed to enhance the girls' sense that having CAH was something bad and, thereby, sensitized them to stigma experiences and anticipation (“stigma coaching”; Schneider & Conrad, 1980).

In general, stigmatized groups in society can be categorized as suffering from either visible stigma (e.g., people with obesity) or concealed stigma (e.g., people with a homosexual orientation) (Pachankis, 2007). Girls and women with CAH are likely to suffer from both. Our results show that stigmatization by others and its anticipation by women with CAH (as implied by avoidance) was primarily associated with easily noticeable features that are characteristic of the CAH condition, especially when a woman's androgen levels are not well controlled: hirsutism of face, limbs, and body, deep voice (in adults), stocky or masculine body built, obesity, reduced breast development, and masculinized genitalia. Genital atypicalities are exposed at most to only a few persons in the environment: to parents and siblings when patients are young, to romantic/erotic partners at later ages, and to

the patients' doctors at all ages, but they are usually hidden from other people in the general social environment. It is, therefore, not surprising that stigma related to genital ambiguity was rarely reported for the specific social contexts on which the present article is focused. Less visible symptoms of CAH such as absence of menses or fertility problems also contributed to the sense of undesired differentness. The concealed-stigma category further includes the status of having a medical condition (CAH owing to 21-hydroxylase deficiency) that requires daily medication and frequent doctor visits. Thus, girls and women with CAH face a combination of coping challenges in regard to both visible and concealed stigma that vary with specific social contexts.

Some of our observations on stigma related to observable features of CAH were quite similar to what has been found about stigma, coping, and (impaired) quality of life in women with polycystic ovary syndrome (Hahn et al., 2005; Himelein & Thatcher, 2006; Jones, Hall, Lashen, Balen, & Ledger, 2011; Keegan, Liao, & Boyle, 2003), and especially in women studied specifically regarding the psychosocial effects of hirsutism (Keegan et al., 2003; Nelson & Nelson, 2010; Sampogna, Tabolli, & Abeni, 2013); both conditions are frequently associated with hyperandrogenemia, but usually without inborn genital ambiguity. Stigma associated with being overweight has also been well documented in non-CAH children and adolescents (Puhl & Latner, 2007) and adults (Brewis, 2014).

From many of the vignettes one gets the impression—and some participants stated so explicitly—that the diverse stigma triggers and occasions combined their psychological impact in some way. Stigma experiences and related anxieties may add up to a general sense of being abnormal in terms of body image or even overall self-image, i.e., become internalized (Link & Phelan, 2001). The emotional impact appears to be enhanced by the fact that many of the features of CAH do not only depart from society's feminine ideal, but may raise doubts about a woman's real gender in a society in which the majority adheres to an ideology of the binary nature of sex and gender. Thus, stigma may become a challenge, or even a threat, to the woman's identity (Major & O'Brien, 2005). The tendency toward internalization appeared to be particularly strong in adolescence, whereas later in adulthood at least some women with CAH appear to develop more of a disregard of the opinions and reactions of others.

The most frequently reported coping mechanism was some form of avoidance: attempts at concealing the visible body features, staying away from situations that involve baring one's body, withholding self-disclosure to others, and presenting a façade of normality. At least in some girls and women, this appears to have led to self-imposed restrictions on social leisure time activities and on peer relationships and friendship formation, both in terms of quantity and quality, which in some may have been complemented by being excluded from peer groups because of their non-feminine appearance.

Given that this study was initiated for the purpose of developing assessment tools for stigma in all conditions of intersexuality, a

significant limitation was its focus on only one syndrome, namely 46,XX CAH. Clinical experience lets us expect that patients with other intersex syndromes are also at increased risk of similar undesired differentness. Yet, patients with CAH need daily treatment with hormonal medications from early childhood on and undergo related medical examinations much more frequently than patients with other intersex syndromes, which may enhance stigma consciousness. On the other hand, as most patients with 46,XX CAH are assigned and raised female, at least in resource-rich countries with good diagnostic facilities, many retain fertility potential and also do not have to deal with the problem of being raised in a gender that does not correspond to their karyotype and gonadal status, all features that may constitute marked problems for people with other intersex syndromes. Thus, the material generated by the qualitative data from this study constitutes a useful starting point for the development of both the planned assessment method and systematic approaches to assist patients and their parents in coping with intersex stigma. Yet, the CAH-based material needs to be complemented by our clinical experiences with other intersex syndromes or, ideally, by additional formative studies of those, provided samples of sufficient size can be established. Future research will also need to systematically document the variation of intersex stigma with assigned gender, age, race, socioeconomic status of parents, sexual orientation, and cultural/religious gender ideologies as well as stigma resilience and protective factors such as family and social support.

Conclusion

Experiencing, anticipating, and/or internalizing stigma related to their medical condition is quite common among women with CAH; the emotional impact can be intense, and the psychosocial implications pervasive. This conclusion is likely to apply to other syndromes of intersexuality as well. Systematic detailed documentation on samples of sufficient size is still lacking. Tools are needed that are suitable for both research and routine clinical assessment and can, thereby, provide the basis for the systematic development of improved and individually tailored approaches to prevention and intervention.

Acknowledgments The study was supported in part by USPHS Grant HD-38409 and Dr. Khuri by a postdoctoral fellowship T32 MH18264. Susan W. Baker, Ph.D., served as the primary interviewer. We thank all participants for their contribution of effort, time, and data to this study.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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